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Original Communications

MAXIMAL RECONSTITUTION OF THE STENOTIC MITRAL VALVE BY NEOSTROPHINGIC MOBILIZATION (REHINGING OF THE SEPTAL LEAFLET)

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DURING the 9 years which have elapsed since the initial presentation[1] by one of us (C. P. B.) of the first successful modern-type operation for mitral stenosis, many logically to be expected advances have taken place in our understanding of this disease, in our medical and surgical management, and in our operative technique. Members of the medical profession, although skeptical and reluctant at first, have accepted the initially proposed procedure with what has seemed to the innovator to be unbridled and uncritical enthusiasm. This operation has been described under the names "commissurotomy,"[1] "valvulotomy,"[2] "valvuloplasty,"[3] and "valvotomy,"[4] although all of these terms have indicated an essentially identical technique. Those who have raised their voices against the widespread application of the method or its accomplishments, albeit often for reasons which subsequently have been shown to be invalid, gradually have been overwhelmed by the increasing weight of favorable medical opinion.

Today, it is the unfortunate duty of your authors to confirm the over-all conclusion of the dissenters that this is in many respects a disappointing procedure, and to suggest in its stead a more logical and more extensive operative mobilization of the mitral valve based upon a different mechanical principle which we hope and believe will overcome the disadvantages and shortcomings of the older technique. In order to avoid the possibility of confusion with the original operation for mitral stenosis, we suggest an entirely new descriptive

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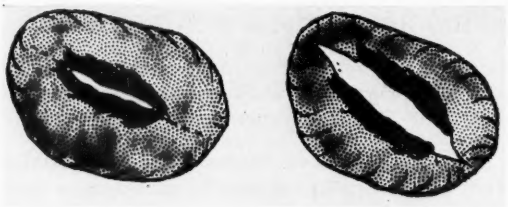
name for the procedure: neostrophingic mobilization of the septal leaflet. This is derived directly from the classical Greek: *neo*—new, and *strophings*—hinge, meaning a new hinging of the septal leaflet.

The original "corrective" operation for mitral stenosis consisted essentially of the insertion of the operator's index finger through an incision made in the tip of the purse-stringed left auricular appendage in order to palpate the stenotic mitral valve orifice, and to split apart the fused leaflets at either or both extremities of the diminutive slit or, failing in this attempt, to effect an instrumental section of the commissures by a valvulotome inserted through the appendage.[4-7] In some clinics, the valvulotome amounted merely to a ring with a cutting edge which was worn upon the finger tip.[8] Some operators have used only a dilating mechanism to accomplish an enlargement of the valve orifice.[9, 10]

In nearly all instances, the surgical principle employed has been that of separating the fused valve leaflets at the commissures, essentially a lengthening of the diminutive valve slit (Fig. 1, *A* and *B*). Appreciating that the most



A. *B.*

Fig. 1.—*A*, Original concept of diminutive stenotic valve slit with fibrosed (in black) margin completely surrounded by flexible valvular tissue (shaded).

B, Original concept of simple linear lengthening of the valve slit by incision or splitting at each extremity. It was felt that once the marginal tissue had been divided well through the scar and into flexible tissue, a great portion of the natural "flutter-valve" action would be restored without the creation of any regurgitation. (*From* O'Neill, Glover, and Bailey: Commissurotomy for Mitral Stenosis, J. Internat. Coll. Surg. 8: 359, Fig. 7, 1950.)

basic (although this admittedly is an oversimplification) action of the normal mitral valve is that of the flutter valve, cardiac surgeons were confident that they were restoring, in inverse proportion to the extent of intrinsic pathologic change, a portion of the normal-type valve action. With the valve so mobilized, little or no regurgitation is created. Since even a rather slight restoration of valvular function may suffice to bring about great clinical improvement in patients with mitral stenosis, surgeons and cardiologists alike were delighted with the immediate benefits. They were not seriously disturbed by the usual persistence of typical auscultatory evidences indicative of residual mitral stenosis.[11-13] Nor were they concerned with the likelihood of the development of recurrent mitral stenosis,[5, 14-16] although examination of typical pathologic specimens indicated that the severely stenotic mitral valve had become converted essentially into a fibrous stricture (Fig. 2).

The frequent reinforcement of the primary leaflet fusion by subvalvular changes[4, 8, 17, 18] largely has been ignored although it is very evident that even

completely mobilized leaflets cannot become separated freely during diastole if they continue to be bound together by cross-fused chordae or by a directly adherent papillary muscle (Fig. 2).

With the passage of time, certain significant modifications were added to the original operative technique. These included "flush-out" of the thrombosed appendage[8, 19] and temporary occlusion of the carotid circulation,[8, 20, 21] both techniques having been designed to lessen the incidence of serious arterial embolization. A very important innovation was the "bare-finger" method originally suggested by Husfeldt.[22] The increased tactile sensibility facilitates accuracy of orientation, permits a more effective application of digital disruptive pressure, and ensures critically accurate guidance of any necessary instrumentation.

Fig. 2.—Strictured orifice in a heavily fibrosed valve. (*From* Bailey: Surgery of the Heart, ed. 1, Philadelphia, 1955, Lea & Febiger, p. 515.)

Appreciation of the importance of alterations in the subvalvular supporting structures, as described by Brock,[4] led to the development of definitive methods for relief of any such subvalvular stenosis by Harken[23] and especially by Nichols and Jamison.[24]

The development and perfection of the right-sided approach to the mitral valve[18, 25-28] brought about a revolutionary advance in the technical facility with which the stenotic mitral valve mechanism can be mobilized. At first this procedure amounted merely to a superior type of commissurotomy. It was, however, only a matter of time until the now clearly demonstrable effect of each

of the various steps entailed in the surgery of the stenotic valve would be integrated consciously into a significantly advanced concept for maximal restoration of valvular function.

While Chiechi[29] and others[5, 18, 23, 30] have attempted to describe in detail the movement and function of the various components of the mitral valve mechanism, for the purpose of simplification and clear surgical understanding, the major mitral valve leaflets may be considered to act in the same manner as the

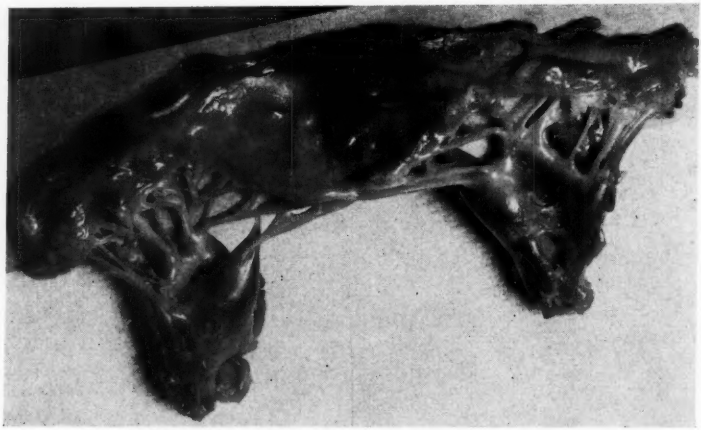


Fig. 3*A*.—The septal leaflet of the mitral valve being shaped like a wedge or a truncated triangle does not permit the two freely separated cusps to become approximated in a straight line. (*From* Bailey: Surgery of the Heart, ed. 1, Philadelphia, 1955, Lea & Febiger, p. 492.)

Fig. 3*B*.—Nearly semicircular line of closure which is characteristic of the normal mitral valve. (*From* Bailey: Surgery of the Heart, ed. 1, Philadelphia, 1955, Lea & Febiger, p. 491.)

two lips of a flutter valve. However, since the septal leaflet is considerably longer from free margin to basal attachment (on the annulus fibrosus) than is the mural, obviously the flutter-valve action must be an eccentric one. Further, largely because of the triangular shape of the septal leaflet (Figs. 3*A* and 3*B*),

the line of valve closure normally is so curved that it constitutes practically a semicircle. This ideal orifice is a far cry from the original surgical objective of simple linear elongation of the valve slit (see Fig. 1). Nevertheless, if the mutually adherent valve leaflets consistently remained thin and flexible, it would be easy enough to accept a slight modification of this concept of linear elongation, the appropriate line of separation merely being extended in arcuate fashion. It must be admitted that in certain lightly fused valves with but slight fibrous change, such a mobilization will accomplish a nearly ideal mechanical result, the original flutter valve being functionally re-established.

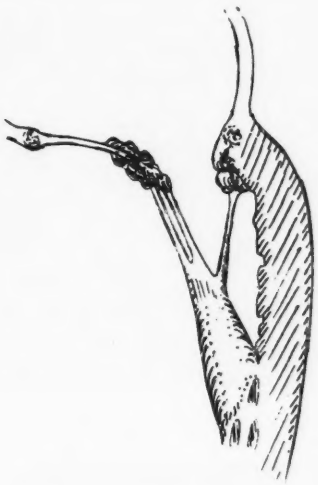


Fig. 4A.—Diagrammatic illustration of equal extents of involvement of both leaflets by the rheumatic process. The distal third of the septal, but the entire apicobasal length of the mural leaflet are thickened and fibrosed. Contraction of the chordae of the third order may draw the shrunken mural cusp against the ventricular wall.

However, in actual practice, the finding of such a minimal degree of pathologic change in mitral stenosis is exceptional. Usually the entire mural leaflet is shrunken from free margin to base, is thickened and fibrosed—perhaps infiltrated with calcific salts. This involvement may extend equally for the entire annular and marginal length of the mural leaflet or it may be limited or concentrated toward one pole of the valve. Sometimes the pathologic change involves chiefly one of the original valve commissures—more often the "posterior." However, in most patients who come to surgery the majority of the length of the mural leaflet is so carnified, so shrunken, or so retracted against the ventricular wall that it is inconceivable that any worth-while mobility could be imparted to it. In all probability the maximal functional value of this altered leaflet lies in the fact that it presents an immobile ridge or shelf extending into the atrioventricular channel against which the surgically mobilized septal leaflet can abut to provide valvular competence. Undoubtedly this rigid projecting shelf impedes to some degree atrioventricular blood flow. Because of the relatively large size of the total atrioventricular passageway, this impedance

must be slight. For the sake of surgical thinking, the junctional tissues[29] may be considered to be part of the length of the mural leaflet since they are subject with it to a similar process of deformation.

Paravisini-Parra[31] has postulated that the mural leaflet of the mitral valve is more susceptible to the ravages of the rheumatic process than is the septal and it is true that extreme degrees of pathologic involvement and retraction

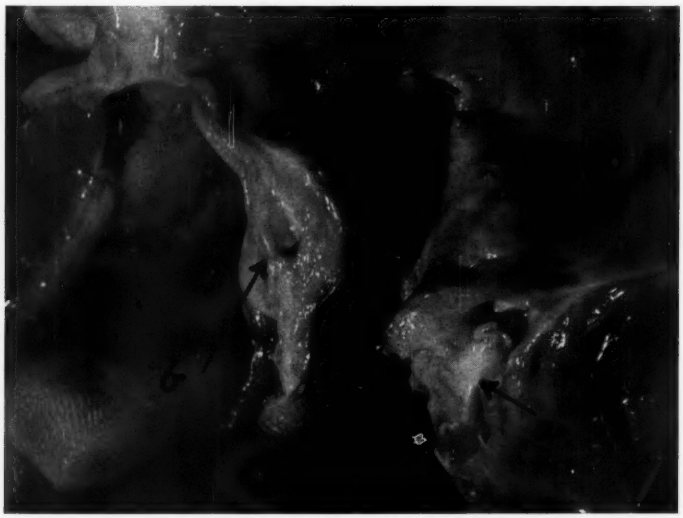


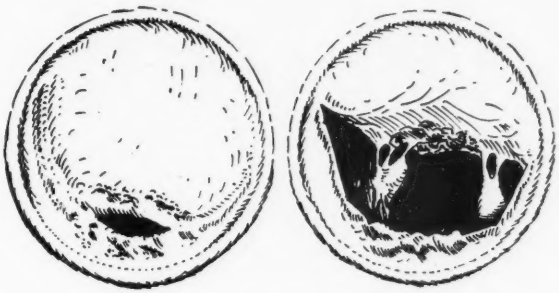
Fig. 4*B*.—Photograph showing retracted carnified mural leaflet (*a*) and fibrosis of the distal portion of the septal leaflet (*b*). Note thin flexible basal portion of latter.

Fig. 4*C*.—Similar photograph of a specimen of mitral stenosis in which the mural cusp is not retracted but in which the characteristic disproportion in relative per cent of involvement of the respective cusps is shown. Note thin flexible proximal portion of septal leaflet.

do appear to be more frequent in this leaflet than in the septal. However, this may be more apparent than real because of anatomic differences in the respective structures of the two leaflets. The chordae of the third order attach only to the

mural leaflet. When they become thickened and contracted due to the organization of inflammatory exudate accumulated upon their surfaces and within the angle formed by the junction of the ventricular aspect of the mural leaflet with the inner endocardium of the left ventricular wall, they may draw the mural leaflet toward or even against the ventricle (Fig. 4*A*). In extreme instances, this retraction may cause regurgitation because it can amount functionally to an absence of the mural leaflet over a given area. In most cases the leaflet continues to project as a rigid though diminutive shelf.

An equal extent of rheumatic involvement of both leaflets from the free margin basally will carnify (or calcify) only one third of the septal leaflet while an equivalent amount of pathologic change will render practically the entire apicobasal length of the mural leaflet completely rigid. In other words, rheumatic thickening and fibrosis of the septal leaflet nearly always is limited to the distal (marginal) third, the middle and basal thirds remaining thin, flexible, and grossly free of disease (Figs. 4*B* and 4*C*). This observation is basic to the concept of "rehinging" the septal leaflet (neostrophingic mobilization). No



A. *B.*

Fig. 5.—*A*, Diagrammatic representation of characteristic findings in severe mitral stenosis. Note total fibrosis of central portion of the mural leaflet.

B, Extensive mobilization of the septal cusp from the rest of the (continuous) valve ribbon so that the cleavage extends into fully flexible tissue at either pole provides free hinging upon its middle third, unless restricted by cross-fusion of the subvalvular supporting structures.

matter how rigid or calcified the septal leaflet may seem on first palpation, complete mobilization from the mural leaflet (and the junctional tissues and/or commissural leaflets when present) to a point at either extremity which is beyond any gross thickening or disease (usually slightly beyond the original location of the commissures) will enable the septal leaflet to bend freely upon its middle uninvolved zone unless restricted by cross-fusion or shortening of the chordae tendineae or by a direct adherence of one or both papillary muscles to the valve apex (Fig. 5, *A* and *B*). While we have recognized such direct papillary attachment in only 30 per cent of stenotic valves at the time of surgery, it is readily apparent to the examining finger that a considerable restriction in the degree of possible leaflet divergence is present at one or both poles in more than half of all operated cases (Fig. 6, *A* and *B*). Usually this is due to a shortening of the normal "yoke" of suspension created either by shortening of the chordae or by an apical lengthening of the valve leaflets due to accretion of organized exudate as described by Brock,[5] or to both.

Only by extensive longitudinal splitting of the restricting pathologic chordopapillary complex (when present) can this binding or limitation of valve movement be abolished! In most instances, this will necessitate complete longitudinal splitting of the papillary muscle (perhaps initiated by direct incision at the "crotch" of the suspension) fully to its ventricular origin. Fortunately, such longitudinal separation of the papillary muscle does not influence its ability to support the respective leaflets and, hence, does not tend to diminish valvular competence (Fig. 6, *C* and *D*).

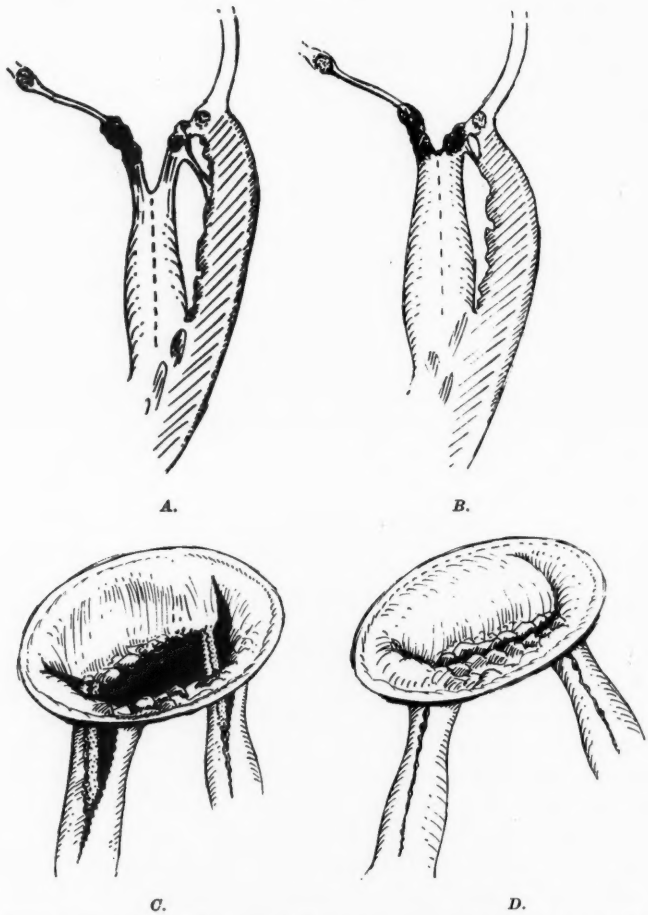


Fig. 6.—*A*, Diagrammatic illustration of shortening of the normal "yoke" of suspension of the mitral valve. This restricts possible leaflet divergence. It may be caused by actual shortening of the fibrosed chordae or by "lengthening" of the leaflets due to organization of exudate deposited upon their free margins.

B, Direct adherence of chordopapillary fusion mass to valve apex due to extreme stage of this process.

C, Complete rehinging of septal leaflet by extensive longitudinal splitting of the papillary muscles as well by total mobilization of this leaflet from the rest of the valve ribbon.

D, Even though the mural cusp is rigid and immobile, free flap-like movement of the septal leaflet provides excellent valve action and full competence.

Once the septal leaflet has been mobilized from the rigid mural leaflet and junctional tissue, the separation being extended fully to its flexible mid-portion, providing any subvalvular restriction has been eliminated, it will swing freely like a great sail in response to the hydrodynamic stresses natural to this region. Its free margin diverges from the atrioventricular channel during ventricular diastole and returns during systole to abut against the passive mural ridge,

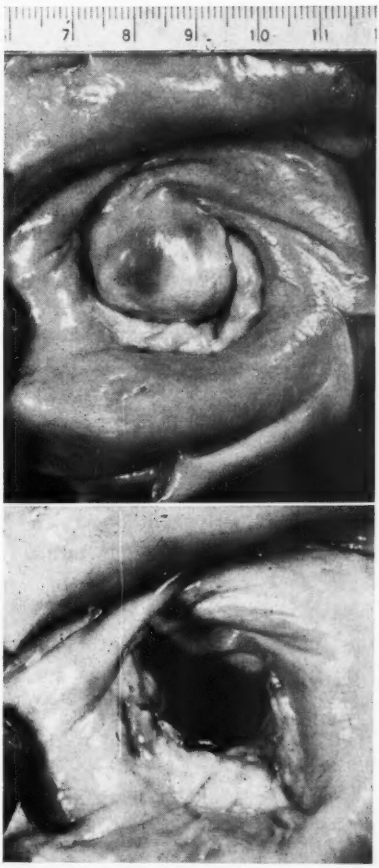


Fig. 7.—*A*, Atrial view of stenotic mitral valve after neostrophingic mobilization (closed position).
B, Same valve in opened position. Note that the fibrosed mural leaflet moves little if at all.

providing competence! Thus, the "flutter-valve" action of the normal bicuspid mitral valve is replaced by a "flap-valve" action of the now nearly unicuspid mitral valve,[8] a characteristic mechanism in certain other vertebrate species (notably the turtle). While such modified valve action obviously is not entirely normal to the human subject, it is, nevertheless, an efficient functional

mechanism. Moreover, it is one which can be established by surgery in most patients with mitral stenosis, whereas a normally functioning "flutter-valve" can be restored but rarely (Fig. 7, *A* and *B*). In those relatively rare instances of very slight deformation, a similar type of surgical mobilization merely results in an exquisitely perfect normal type of bicuspid valve action since, in actuality, it then amounts to a complete renovation of the valve.

DISADVANTAGES OF PREVIOUS OPERATIONS

While the objections to the former method of commissurotomy apply with especial force to those procedures which have been performed through the disadvantageous left-sided approach, they also are pertinent in lesser degree to similar operations performed from the right. It was as much limitation of the earlier concept which restricted the surgeon's operative initiative as the technical faults in his approach.

The fundamental criticisms are the following:

1. Because of technical and conceptual limitations, the extent of valvular mobilization achieved by efforts at commissurotomy by the older method usually amounted to but 20 to 30 per cent of the ideal. In two thirds of the patients, but one commissure was opened.[8, 18] It is believed that the per cent of functional restoration usually has been significantly low. This has been attested by the consistent persistence of auscultatory manifestations of mitral stenosis after surgery as well as by physiologic evidences of residual obstruction at the valve level.

In many instances this residual stenosis was reinforced and aggravated by an element of subvalvular stenosis, usually unrecognized and less often relieved (now believed to be present to a significant extent in more than half of these patients).

2. Inadvertent production of significant mitral regurgitation. This was a logical and not infrequent end-result of the customary employment of the clumsy and inefficient left-sided approach and the uncertain orientation which it affords.

3. Unsatisfactory clinical course of operated patients. While it is felt that over 75 per cent of patients receive significant benefit from a mitral commissurotomy operation, in many cases this has amounted merely to prolongation of a life of invalidism rather than to the rehabilitation which an ideal correction of the mitral obstruction might have offered.

4. The increasing evidence that mitral commissurotomy is attended by a high incidence of recurrence of mitral stenosis after the elapse of several years.[16] It is believed that 20 to 40 per cent of patients operated by the older technique will suffer from restenosis and ultimately will require repetitive surgery.

5. A seemingly irreducible incidence of arterial embolization due chiefly to dislodgment of thrombotic fragments from a clotted left auricular appendage.[8, 27]

RESULTS OF NEOSTROPHINGIC MOBILIZATION

Because of increasing dissatisfaction with even the improved commissurotomy procedure possible by the right-sided approach,[26, 27] the described new concept of a method for complete reconstitution of valve action was born.

In spite of the evolution of this concept of rehinging of the septal leaflet which would seem to approach the surgical ideal, we have not always succeeded in accomplishing this objective in practice. Among 227 consecutive patients who have been operated for essentially isolated mitral stenosis at our clinic since the deliberate application of this principle surgically for the first time on Sept. 26, 1955, it is felt that a completely satisfactory mobilization was accomplished in only 70 per cent (Table I). In 20 per cent, the technical achievement (although less than desired) was considered to be equal or superior to that which would have been produced in the same individuals by the older technique of commissurotomy. In most instances the failure was related to termination of the mobilization prior to logical completion because of the beginning development of a regurgitant jet at one or both commissures.

TABLE I. ESTIMATES OF ACCOMPLISHMENT IN RESTORATION OF THE MITRAL VALVE, BOTH ANATOMICALLY AND FUNCTIONALLY

Anatomic opening 100%, function 100%	50	156	70%
More than 90% of valve function restored	106		
Acceptable (over 70% of function re-established)		23	10%
Unsatisfactory results		48	20%
Limited valvular opening	24—10%		
Increased or created regurgitation	19— 8%		
Subvalvular fusion not completely relieved	5— 2%		
Total	227 Cases		

Instrumentation was employed in 181 patients (Table II).

TABLE II. PROPORTION OF DIGITAL AND INSTRUMENTAL MITRAL SEPARATION

Both commissures opened			218
Left and right knife used	100		
Left knife only used	53	175	
Right knife only used	22		
Digital only		43	
Anterior commissure only			5
Left knife used		4	
Digital only		1	
Posterior commissure only			4
Right knife used		2	
Digital only		2	
Total			227

Regurgitation of greater than 1+ degree was created (or exaggerated) in 19 patients. However, 76 patients have manifested a new systolic murmur postoperatively (Table III). Since many of these patients presented tricuspid disease, this sign is considered of uncertain significance.

Calcification of the valve which was considered severe in 34 instances did not seriously reduce the surgeon's ability to achieve a satisfactory mobilization (Tables IVA and IVB).

TABLE III. PRE- AND POSTOPERATIVE CHANGES IN VALVE COMPETENCE

	PREOPERATIVE GRADE OF REGURGITATION	POSTOPERATIVE GRADE			
		NONE	MINOR	MODERATE	SEVERE
None	167	51	111	3	2
Minor	54		46	7	1
Moderate	6			6	
Totals	227	51	157*	16*	3*

*Seventy-six patients had a new systolic murmur postoperatively.

TABLE IV A. INCIDENCE OF CALCIFICATION OF THE VALVE

No calcification		152	(67%)
Calcified valve		75	
Minimal degree	41		
Moderate degree	23		
Heavily calcified	11		
Total		227	

TABLE IV B. INFLUENCE OF CALCIFICATION OF THE MITRAL VALVE UPON FINAL FUNCTIONAL VALVULAR OPENING

	MORE THAN 90% FUNCTIONAL OPENING OBTAINED		LESS THAN 90% FUNCTIONAL OPENING OBTAINED		TOTAL
Fibrotic noncalcified valve	110	72.4%	42	27.6%	152
Partially or heavily calcified valve	46	61.4%	29	38.6%	75
Totals	156		71		227

TABLE V A. SUBVALVULAR INVOLVEMENT

No subvalvular involvement recognized			99	(43.6%)
Anterior subvalvular changes		28		
Papillary fusion to valve	8			
Chordal agglutination	20			
Posterior subvalvular changes		76		
Papillary fusion to valve	46			
Chordal agglutination	30			
Bipolar subvalvular changes		24		
Papillary fusion to valve	16			
Chordal agglutination	8			
			128	(56.4%)
Total			227	

TABLE V B. THE RESULTS OF TREATMENT OF THE SUBVALVULAR STENOSIS IN THE CASES OF MITRAL RECONSTITUTION*

No subvalvular stenosis	99
Subvalvular changes not treated	
No effect on the valvular function	29
Minimal changes	3
Unable to separate	5
Subvalvular fusion treated	
Complete or almost complete relief obtained	64
Physiologically reasonable function obtained	12
Unsatisfactory separation accomplished	15
Total	227

*Guillotine knife or papillotome used, 12 cases; digital pressure or fingernail used, 84 cases.

Subvalvular pathologic change was found in 128 patients (Table VA). In 70 (30.8 per cent) valves, direct papillary muscle adherence to the valve apex was recognized at one or both poles. However, complete relief of subvalvular stenosis was not accomplished in 20 cases (Table VB).

Arterial embolization was produced at surgery in 7 patients (3 per cent). In 5 patients, this was cerebral; in 2, peripheral (Table VIA). No limb amputations were required. While these accidents were believed to have been produced by inadvertent dislodgment of calcific particles from the valve at the time of surgery, it is possible in some that dislodged thrombotic material from a clotted atrium was responsible (Table VIB). However, avoidance of the left auricular appendage as an entrance route would seem to have reduced the likelihood of thrombotic embolization.

TABLE VI A. INCIDENCE OF ARTERIAL EMBOLISM IN 227 CASES OF MITRAL RECONSTITUTION

	NUMBER OF CASES	DEATHS
Cerebral embolism	5 (2.2%)	2
Peripheral embolism	2 (0.8%)	0

TABLE VI B. RECOGNIZED INCIDENCE OF THROMBOSIS OF THE LEFT ATRIUM

No thrombus felt			204
Thrombus			23 (10%)
Loose fresh thrombus		18	
Required removal	5		
Not disturbed	13		
Placenta-like organized thrombus (not disturbed)		5	
Total			227

The technical results achieved are shown in Tables VIIA and VIIB.

TABLE VII A. OPERATIVE MORTALITY OF OPERATIONS FOR MITRAL STENOSIS

	NUMBER OF CASES	OPERATIVE DEATHS
Mitral commissurotomy from the left side	1,000	77 (7.7%)
Mitral commissurotomy from the right side (before Sept., 1955)	150	13 (8.6%)
Neostrophingic mobilization (from Sept., 1955 to Jan., 1957)	227	11 (4.8%)
Good technical result More than 90% valvular function restored	156	4 (2.5%)
Compromised technical result; Less than 90% valvular function restored	71	7 (10%)

TABLE VII B. THE FOLLOW-UP STUDIES OF MITRAL VALVULAR RECONSTITUTION (MORE THAN 3 MONTHS)

	EXCELLENT	IMPROVED	SAME	DEATHS	TOTAL
Ideal or near ideal functional valvular opening obtained	65 (54.6%)	45 (37.8%)	5 (4.2%)	4 (3.4%)	119 (100%)
Less satisfactory valvular opening established	21 (41.2%)	22 (43.1%)	1 (2.0%)	7 (14.7%)	51 (100%)

The diastolic murmur and sharp first sound of mitral stenosis were abolished (by phonocardiographic determination) in half of the entire series (Tables

VIIIA and VIIIB). By simple auscultatory examination, the rate of apparent abolition of this murmur was much greater (Fig. 8, *A* and *B*).

TABLE VIII A. POSTOPERATIVE DISAPPEARANCE OF MITRAL DIASTOLIC MURMUR AFTER NEOSTROPHINGIC MOBILIZATION (PHONOCARDIOGRAPHIC DETERMINATION)

	NUMBER OF CASES	DISAPPEARANCE OF DIASTOLIC MURMUR	
Complete or near complete restoration of valvular function	148	75	(51%)
Incomplete restoration of valvular function	56	25	(45%)
Total	204	100	(49%)

TABLE VIII B. POSTOPERATIVE DISAPPEARANCE OF THE SHARP FIRST MITRAL SOUND AFTER NEOSTROPHINGIC MOBILIZATION (AUSCULTATORY DETERMINATION)

	NUMBER OF CASES	DISAPPEARANCE OF SHARP FIRST SOUND	
Complete or near complete restoration of valvular function	119	55	(46%)
Incomplete restoration of valvular function	44	23	(50%)
Total	163	78	(42%)

A.

B.

Fig. 8.—*A*, Phonocardiographic record of heart sounds at apex prior to neostrophingic mobilization of stenotic mitral valve in Patient C. A. (Preoperative.)

B, Postoperative (3 weeks) phonocardiographic study in Patient C. A. showing complete abolition of the diastolic murmur.

It is interesting that there is no direct linear relationship between the functional restoration of the valve and the old measurement of valvular opening in "fingers."

TABLE IX. RELATIONSHIP BETWEEN ESTIMATE OF PERCENTAGE OF FUNCTION AND OPENING MEASURED IN "FINGERS" IN CASES OF NEOSTROPHINGIC MOBILIZATION

	MORE THAN 90% FUNCTIONAL OPENING		LESS THAN 90% FUNCTIONAL OPENING	
More than 2 fingers	16	10%	4	5.6%
2 fingers	86	55%	21	30%
1 and ¾ fingers	47	31.4%	29	40%
1 and ½ fingers or less	7	3.6%	17	24.4%
Totals	156	100%	71	100%

THE SURGICAL TECHNIQUE

The Thoracic Entrance.—The patient is placed in the supine position. A right anterolateral thoracic incision is made from the sternal border to the mid-axillary line, curving below the breast in the female.

The pectoral muscle fibers are split laterally and incised medially to expose the fourth intercostal space. The intercostal muscles and pleura are divided extensively from the internal mammary vessels to the transverse processes of the vertebrae. The former are divided between individual ligatures. The fourth and fifth costal cartilages may be divided at their sternal junctures. The ribs are spread widely apart in gradual increments by a Finochietto type of fixed retractor in order to avoid fracture. The right lung is compressed posteriorly.

The Pericardial Incision.—The pericardial sac is incised parallel and 1.5 cm. anterior to the right phrenic nerve for its entire length. The caudal extremity of this longitudinal incision may be extended anteriorly to the sternal border to obtain additional exposure.

The vessels which cross the incised portion of the pericardium are individually ligated and the lips of the incision in this sac are retracted with weighted stay sutures.

The Cardiac Incision.—The interatrial groove is easily identified by gentle upward retraction of the right atrium by the gauze-covered fingers of an assistant. It will be seen, at least during certain phases of the cardiac cycle, as a distinct line or depression communicating the posterior aspects of the inferior and superior venae cavae.

The mid-portion of the groove is dissected to a limited extent, usually 1 cm. in length and 8 mm. in depth, but with individual variations.

A purse-string suture of heavy No. 2 braided nylon, swedged on a curved round-edged needle, is placed widely about this dissected area. The line of encirclement should be at all points at least 1 cm. distant from the line of dissection. Each bite of the suturing needle should penetrate into the corresponding atrial lumen. The external loops of the suture should be made longer than the internal ones. The free ends are incorporated within a Rumel-Belmont tourniquet.

In order to prevent unexpected separation of the interatrial septum from taking place during the left atrial exploration, a 3-0 cotton stay suture is placed at either extremity of dissection to "hold" the interatrial septum together. The ends of these sutures are kept long for usage at the time of closure of the cardiac wound.

A suture of No. 2 braided nylon swedged upon a large curved needle is passed through both lips deeply beneath the line of dissection, starting at the right atrium. It traverses the septum to emerge from the left atrium and is returned at a distance of 1 cm. to form a large mattress suture. Both ends of this mattress suture are drawn upward and are loosely attached to the drapes in order to prevent subsequent entanglement. At the conclusion of the valvular surgery, this suture will be tied down to evert (Fig. 9) the lips of the incision in the left atrial wall. A pair of Morse* scissors (with points and outer edges of blades sharpened) is laid closed against the palmar surface of the bare left index finger which has been washed with saline and dipped into a dilute

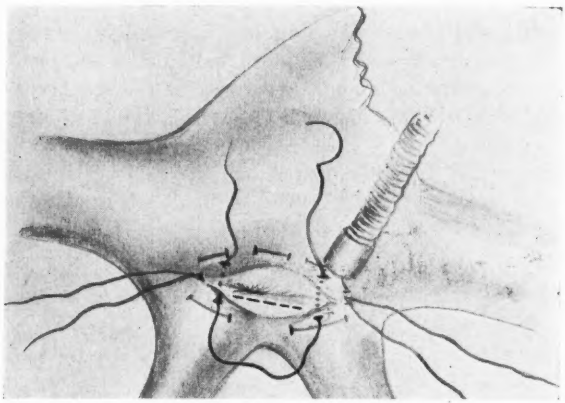


Fig. 9.—Dissection of interatrial groove and placement of various controlling sutures. Dotted line shows site of proposed incision into left atrium.

heparin solution (1:1000). The finger and scissors are inserted into the dissected portion of the interatrial groove and the scissors are advanced to puncture the exposed septal aspect of the left atrial wall. Then the blades are fully opened to produce an adequate entrance, but one which readily can be tamponaded by insertion of the index finger. As the blades are withdrawn in the partially opened position, a small gush of arterial blood will escape before the tip of the advancing index finger occludes the opening completely. The finger enters the atrial chamber slowly with a gentle twisting motion, the opening gradually becoming dilated to fit its increasing circumference.

Exploration of the Left Atrium.—The finger tip soon reaches the mitral valve orifice. If the left atrium is enlarged considerably, it may be necessary to invaginate the atrial wall partially in order to reach the valve.

*Available from G. P. Pilling and Sons, Philadelphia, Pa.

A complete and thorough examination of the left atrium should be carried out in all cases in order to rule out the possibility of a coexisting septal defect, anomalous pulmonary venous drainage, or the presence of a thrombus or intracardiac tumor before the definitive steps of the valvular surgery are undertaken.

Intra-atrial Thrombus.—Intra-atrial thrombosis was found in 10 per cent of this series. If the thrombus is securely adherent to the atrial wall, it should be left undisturbed, extraordinary gentleness being exhibited during the entire course of cardiac manipulation. In such cases, it is our custom to start anticoagulant therapy (first with heparin and later with Dicumarol), beginning 12 hours postoperatively, and continuing for about 2 weeks in order to lessen

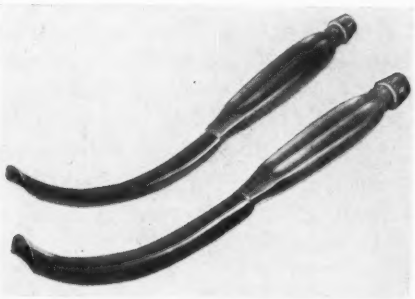


Fig. 10*A*.—Litwak suction tips.

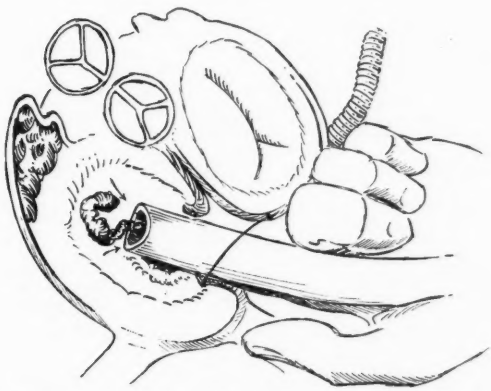


Fig. 10*B*.—Method of aspirating floating or very loosely attached thrombotic material from left atrium by passage of suction tip into heart along the palmar surface of the intracardiac finger.

the likelihood of propagation of the thrombus. If the clot is loosely attached to the atrial wall or floating freely, it may be removed by utilizing one of the Litwak suction tips which may be passed into the atrial chamber along the palmar surface of the intracardiac finger (Figs. 10*A* and 10*B*).

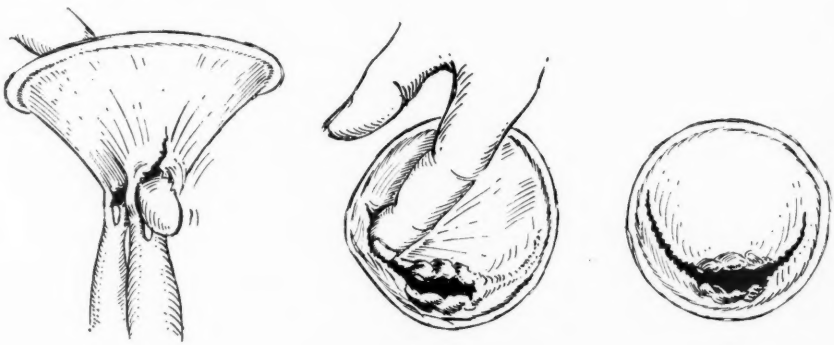
Examination of the Valve.—The finger tip is held poised over the valve orifice at a distance of 1 cm. in order to detect a possible regurgitant jet. If noted, its exact localization is determined and an estimate is made of its severity. In carefully selected cases such as in this series, such a jet should never exceed a one or two plus magnitude and nearly always will be the former. Usually it will be felt to be located at the central portion of the slit-like opening.

The atrial aspect of the valve is palpated to determine the size and configuration of the mitral orifice and to recognize the possible presence of calcification.

In most instances, the valve slit will not admit a finger tip and will be centrally located with respect to the annulus fibrosus which is readily discernible from this approach.

If calcification is present, its type and amount, and especially any encoachment toward the annulus fibrosus, should be determined carefully.

Before attacking the valve, the operating table should be tilted in order to elevate the left side of the patient by 20 to 30 degrees. In this posture, the alignment of the extremities of the residual valve slit will be fitted nicely for instrumentation by either the right or left valvulotome, should such be needed. However, it is always our practice first to attempt to separate the valve structures by digital pressure.



A. *B.* *C.*

Fig. 11.—*A*, The splitting of the diminutive valve slit may be started by simply forcing the finger through the opening, or (cautiously) by application of lateral pressure.

B, The split is extended by lateral or "paper-cutter" digital pressure until the septal mobilization is complete at this pole.

C, An attempt is made to develop a similar extensive line of separation of the opposite valve pole.

MOBILIZATION OF THE VALVE

Digital Separation of Fused Valve Leaflets.—The intracardiac finger is passed forcibly through the diminutive valve slit. In favorable cases, the fused valve leaflets will begin to separate at one or both commissures. Should this occur, an attempt will be made to increase this by intermittent passage of the finger over the apex of the slit in the direction of the ventricular apex in a "paper-knife" fashion. Direct lateral or end-on pressure against the apex of the split may also be tried, always cautiously, however (Fig. 11, *A* and *B*).

In most cases, persistence and repetition in application of this finger pressure soon will develop a long upward "horn" at the extremity of the horizontal portion of the valve slit. The tip of this "horn" will come to run almost parallel with the annulus fibrosus, usually at a distance of 3 to 5 mm. from it, but sometimes practically along its margin. Obviously, this latter type of cleavage is the result of deformation and retraction of the valvular ribbon since the orifice normally does not approach the annulus. Careful examination of the apex of the split after each stroke of the finger will reveal even the slightest new increment of regurgitation. This will be felt as a localized high velocity systolic jet if small, or more diffuse, involving a greater portion of the aperture, if it is larger. Should such a jet appear at the extremity of the valve slit, it is probable that the splitting has deviated from the proper line of cleavage. No further attempt at separation of that pole of the valve is then permissible lest it lead to the creation of a serious or even lethal amount of insufficiency.

After commissural separation has been completed at one pole of the valve, at attempt should be made to split the opposite one (Fig. 11, *C*).

Instrumental Separation of Fused Valve Leaflets.—Although digital mobilization of the valve leaflets is more efficient and feasible by the right-sided route than from the left, many valves still will require definitive instrumentation. For the right-sided approach, it has been necessary to modify our original right and left guillotine knives, both with respect to length and to the arc of curvature.[26, 27] Further, the principle of the Nichols papillotome[24] has been incorporated into the design.*

The anterior lip of the dissected interatrial groove is picked up with a pair of Nelson forceps and the appropriate closed guillotine knife is passed into the left atrium along the palmar surface of the operator's left index finger.

It is our policy first to incise the more favorable valve extremity (unless it already has responded to digital pressure). Often this will be the anterolateral (really left). The left-sided guillotine knife is passed through the valve opening and is brought to bear against the apex of the left extremity of the nearly transversely directed valve slit. As the jaw is opened by retracting the sliding blade, the under-beak will slide beneath the valve margin.

Digital palpation of the chordopapillary supports now is indicated to establish whether the under-beak of the instrument has been placed properly between the two parallel rows of supporting chordae or possibly through the mid-portion of a chordopapillary fusion mass. It may be necessary to rotate the instrument slightly, taking care that it does not slip from its position in order to ensure a horizontal incision at this valve extremity. By compressing the handles, the upper blade of the instrument is caused to cross the lower, shearing through 4 mm. of scarred or calcified marginal valve tissue. The closed instrument is withdrawn slightly while the finger tip remains poised momentarily over the site of section to detect even the slightest change in valve competence.

Usually it is wise to wait a few moments for reassurance from the anesthetist that the patient's condition has remained at, or has been restored to, a satisfactory level.

*Obtainable from Storz Instrument Co., St. Louis, Mo.

Now, an attempt should be made to split the partially divided commissure with the intracardiac finger tip. If this succeeds, no further instrumentation will be necessary and the guillotine may be removed from the heart. However, it may be found necessary to use the knife one or more additional times. In such a situation, the valvulotome should be readvanced and re-engaged at the new valve "apex." This time, the knife must be angulated upward at 30 degrees from the horizontal line and the new bite be taken in this direction.

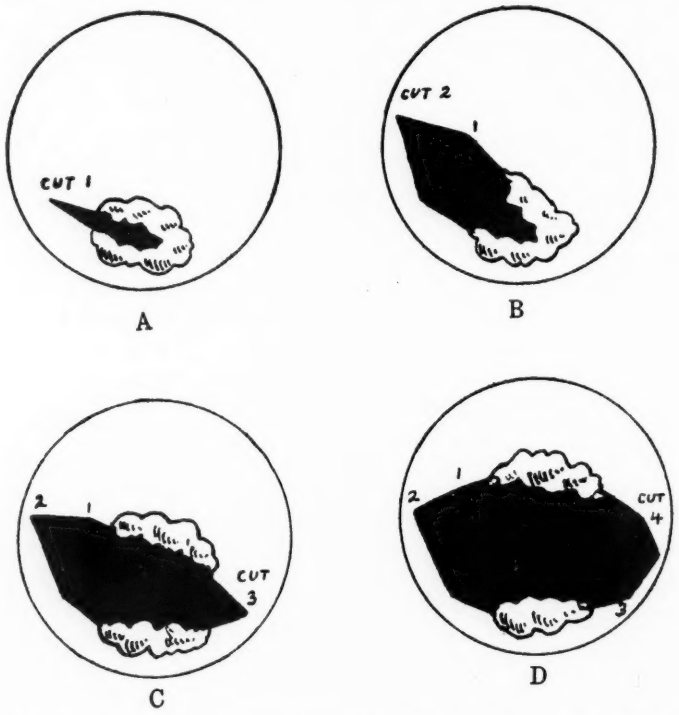


Fig. 12.—*A*, Diagrammatic representation of nearly horizontal section accomplished by first bite of guillotine against the anterolateral (really left) commissure.

B, Second (and perhaps even a later) bite is directed at an increasing upward angle in order to follow the natural curve of the line of valve closure. Shaded area indicates increase in size of orifice.

C, First bite against the posteromedial (really right) commissure is directed horizontally.

D, Any additional instrumental division of this pole must be guided in an increasingly upward fashion. Note extensive increase in effective valve area (shaded portion).

If still another bite should be required, this should deviate further upward from this line (almost 60 to 90 degrees from the original line). At the tip of a well-formed horn, the line of section is nearly vertical to the mid-portion of the valve slit.

If digital pressure becomes effective at any time, the valvulotome should be withdrawn completely from the heart and the separation continued digitally.

As soon as a satisfactory (or compromised because of regurgitation) opening of the anterolateral pole of the valve is obtained, the operator's attention

will turn to the posteromedial (really right) pole of the valve. This time, the right-sided guillotine knife will be used in much the same manner described for the left-sided one.

However, it is slightly more difficult to locate the proper line of separation anatomically or functionally since the extremity of the valve may be rounded rather than angular, curves less steeply upward than the anterolateral one. Again, digital localization of the subvalvular supporting structures will guide the surgeon.

The same precautions should be taken not to deviate from the proper line of separation (which again is more functional than anatomic) (Fig. 12, *A*, *B*, *C*, *D*).

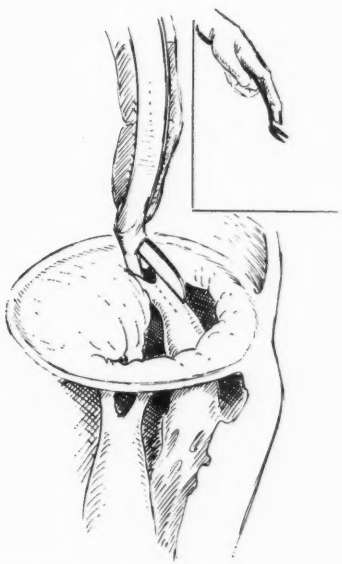


Fig. 13*A*.—The longitudinal separation may be started in difficult cases, especially those with a fibrosed "short yoke" by dividing the "crotch" with a specially designed pair of scissors.

The Treatment of Subvalvular Changes.—In every case after valve separation is deemed to have been obtained as fully as feasible, a search should be made for an element of subvalvular obstruction. If such is found, removal of this obstruction is essential to complete renovation of the valvular mechanism.

An attempt should be made to separate every abnormal chordopapillary fusion whether the papillary muscle is directly adherent to the valve apex or whether a "short-yoke" chordopapillary mechanism is involved. Usually, direct pressure with the finger tip against the top of the mass will suffice to start a longitudinal splitting of the muscle mass which may and should be extended fully to the ventricular attachment. If the atrial extremity of this papillary mass is unusually resistant, it can be divided by a pair of special papillary

scissors* which are designed to be passed into the heart along the palmar aspect of the intracardiac finger. Thereafter, splitting usually will be accomplished easily (Figs. 13*A* and 13*B*).

Closure of the Cardiac Wound.—After full (or compromised) neostrophingic mobilization of the mitral valve has been completed, the finger is withdrawn slowly from the opening in the wall of the left atrium, tamponading the wound surface as the initially placed mattress suture is finally tightened. Usually this will establish complete hemostasis. Should it fail, tightening of the purse-string suture will supplement it. If there still is persistent bleeding, the two lips of the dissected interatrial groove may be approximated with Nelson forceps while they are being sutured.

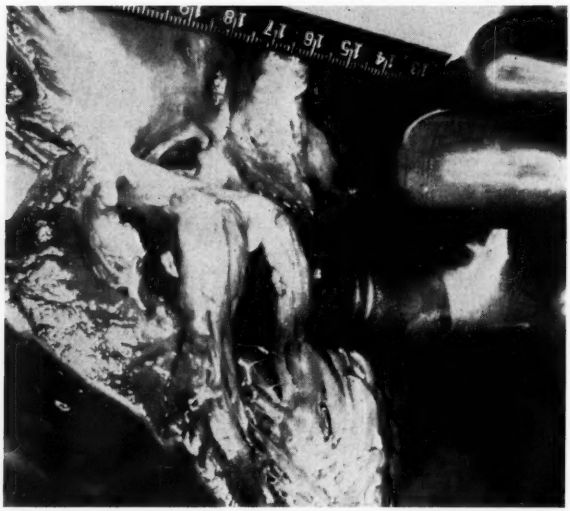


Fig. 13*B*.—Autopsy performed operation showing fully mobilized septal leaflet, both directly adherent papillary muscles having been split to their ventricular wall attachment.

The dissected groove is repaired with one continuous horizontal everting suture and one re-enforcing simple suture using the long ends of the previously placed 3-0 cotton stay sutures.

After thorough flushing with normal saline, the pericardium is closed tightly enough to prevent herniation of the heart to the right but loosely enough to permit free drainage of any accumulated exudate into the chest cavity.

Two multiwindowed catheters are placed intercostally for postoperative drainage and are connected with a water-seal drainage bottle. The ribs are approximated with pericostal sutures of No. 2 double chromic catgut. The muscles and fascia are closed with continuous No. 1 chromic catgut sutures. Subcuticular No. 30 steel wire is used for skin closure.

*Obtainable from the Storz Instrument Company, St. Louis, Mo.

Exploration of the Tricuspid Valve.—Routinely, the tricuspid valve should be explored[18, 26, 27] by inserting the finger through an incision made into the tip of the right auricular appendage if the patient's general condition permits this step. Since, as shown previously, one fourth of all the cases of mitral stenosis have a certain degree of tricuspid valvular involvement and 8 per cent require definitive surgical correction of stenosis, it usually is worth while to spend another extra ten minutes of operating time in examining this valve.

OPEN HEART SURGERY FOR MITRAL STENOSIS

Obviously a similarly extensive type of valvular mobilization might be accomplished by an open technique using one of the existing pump-oxygenator combinations. Here, too, the use of the right-sided approach is essential, but the left atrium must be incised longitudinally for a greater distance. Usually the incision is made just dorsal and parallel to the interatrial groove.

However, because the appropriate line of valve division is not strictly an anatomic but rather a functional one, with an open technique the operator is at a distinct disadvantage in that he is unable to appreciate the earliest stage of creation of incompetence of the nonfunctioning valve. While one might think that the proper line of commissural obliteration would be clearly apparent to visual inspection, it often is most confusing to decide upon its location. Because of the appreciable risk of creating inadvertently a serious grade of insufficiency which cannot become evident until full cardiac action has been restored (and therefore is essentially irremediable) the operator will tend to restrict the extent of valve mobilization to less than the logical ideal. Similarly, he may be loathe to split the papillary muscles fully to the ventricular wall.

For these reasons, although with a personal open surgical experience admittedly limited to but 3 patients with this disease, we have felt that the expert operator will perform a better mitral valve mobilization with the simpler and safer closed technique than with the open.

SUMMARY AND CONCLUSIONS

After a 9-year clinical experience including more than 2,000 patients subjected to mitral commissurotomy through the standard left-sided approach, and close to 500 operated during the past 4 years by the right-sided method, a new and technically much superior type of valve mobilization has been evolved. It is based upon the principle of converting the irretrievably destroyed flutter-valve action of the mitral valve to an efficient flap-valve mechanism.

This effect is accomplished by a rehinging or new-hinging of the septal cusp not only through its flexible mid-zonal tissue, but also with respect to the usually shortened papillary suspension yoke which must be split longitudinally. Such a maximally corrective procedure has been described under the term "neostrophingic mobilization" of the valve. It cannot routinely be accomplished except with the technical advantage of the right-sided approach.

It has been shown that most patients with mitral stenosis have received but partial (as shown by the persistence of auscultatory signs) and often

temporary relief of their valvular obstruction when the older left-sided commissurotomy procedure has been carried out. Restenosis ultimately may be expected in a large proportion of these patients who have, in actuality, been subjected merely to linear (and limited) division of a fibrous strieture. It is believed that recurrence of mitral stenosis after complete neostrophingic mobilization will be rare indeed since the operation amounts actually to construction of a valve mechanism of different type, and since all the tissue cleavages extend well into normal unfibrosed structures.

The early operative mortality and morbidity associated with complete reconstitution of the valve is distinctly less than that associated with less complete operations (see Tables VIIA and VIIB).

Because of certain unavoidable handicaps experienced during attempts to carry out an extensive mobilization of the valve structures with maintenance of full competence when the open technique is employed (using the pump-oxygenator), it is felt that a better and safer reconstitution of the stenotic mitral valve can be achieved by the closed method.

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SURGICAL TECHNIQUE FOR REPAIR OF HIGH VENTRICULAR SEPTAL DEFECTS

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THE time-honored requirements for accurate open operation in any region are a well-exposed, quiet, and relatively bloodless field, and a precise and reproducible operative technique. Preoccupation with the problems of whole-body perfusion must not obscure the necessity for the development of adequate techniques for the reparative intracardiac operations thereby made possible.

One of the challenging intracardiac procedures currently performed is the closure of high ventricular septal defects. These are defects located immediately beneath the aortic-valve ring, and thus high in the ventricular septum when viewed from the left ventricular aspect. Approximately 90 per cent of patients having ventricular septal defects who are selected for surgical treatment and who, therefore, have significant hemodynamic derangement, have a ventricular septal defect in this region.

In a previous publication[1] in this JOURNAL, a technique used at the Mayo Clinic for repair of these defects was described, but a footnote to the report indicated it was no longer in use in this institution. Experience with the repair of ventricular septal defect, either alone or as part of the tetralogy of Fallot, has led to gradual modifications in technique. The method herein described has been used since the adoption of purposeful cardiac arrest and we consider it to be a definitive method.

ANATOMY

Closure of this particular defect requires a clear three-dimensional concept of the compact, complex, and somewhat obscure anatomy of this region.[1, 2] The complexities of the embryologic development of this area of the ventricular septum are beyond the scope of this presentation.[3]

On considering a high ventricular septal defect, such as has been described, from the left ventricular aspect only, the anatomic relationships can be easily comprehended and expressed. The defect presents as an opening that usually equals or slightly exceeds the aortic orifice in diameter. In most instances, the defect is immediately inferior to the right aortic cusp, the adjacent portion of the posterior aortic cusp, and the intervening commissure, and there is no superior remnant of ventricular septum below the aortic ring itself (Fig. 1).

From the right ventricular aspect, however, the region of the defect is partially obscured and rendered relatively inaccessible by adjacent structures,

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including the crista supraventricularis, tricuspid valve, and the papillary muscle of the conus (Fig. 2). Through a right ventriculotomy one perceives that he is first looking into a deep recess, the medial wall of which is formed by the defective ventricular septum. The opposite or lateral wall of this recess appears to be made up, at least in part, of tricuspid-valve leaflet, whereas superiorly the crista supraventricularis narrows the opening into the recess. The opening itself may be bridged by at least one papillary muscle of the conus. At the bottom of this recess is a central fibrous body (or fibrous trigone) to which the aortic-valve ring is firmly attached on the left side, and the tricuspid-valve ring on the right. If one then looks from a caudal point and has proper lighting, he can look into the recess, thence through the ventricular septal defect, and see the aortic cusps.

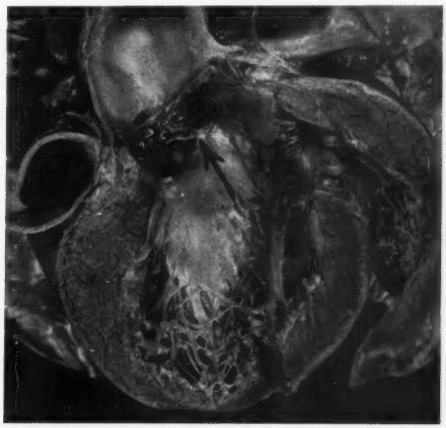

Fig. 1.—Specimen from a 3-month-old boy, showing the ventricular septal defect from the left ventricular aspect. The defect lies inferior to the adjacent halves of the right (*R*) and posterior (*P*) aortic leaflets. The arrow points to the region of the central fibrous body, where the ventricular septum and the aortic, mitral, and tricuspid-valve rings meet. It is essential that the operative repair of high ventricular septal defects begin in this area.

Thus, a striking contrast exists between the appearance of this particular type of high ventricular septal defect when viewed from its right and left ventricular aspects. Indeed, the simplicity of the relationships and the unobstructed view from the left might raise doubt as to the wisdom of approaching the defect through the right ventricle. The impracticability of a left ventricular approach surgically, however, has required persistence in attaining an anatomic orientation from the right, and from this beginning to develop a reproducible technique for complete closure of these defects.

SURGICAL TECHNIQUE

The first requirement for the surgical correction of ventricular septal defects is obviously a means for whole-body perfusion which can be safely maintained for as long as is necessary for the repair. Induced cardiac arrest has clearly become a fundamental necessity to the accurate repair of ventricular

septal defects. The surgical approach is through right ventriculotomy, usually extending from a point just below the pulmonic-valve ring downward nearly to the margin of the heart.

Small cat's-paw retractors are placed at three points about the edges of the ventriculotomy opening, not only to hold the wound open but also to support the heart. Variable degrees of tension on each of these retractors will give variations in the exposure of individual intracardiac regions. The intracardiac

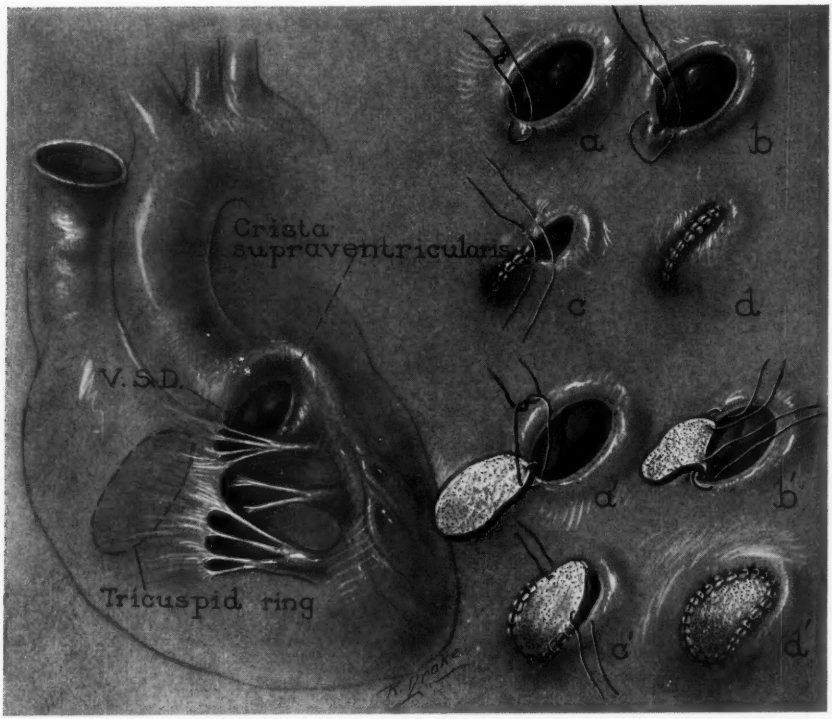

Fig. 2.—Schematic representation of a high ventricular septal defect as viewed from the right ventricular aspect. The insets demonstrate the steps in the surgical repair by both direct suture and insertion of a prosthesis, as described in the text. Note that certain stitches were placed from the left ventricular side, working through the defect, and that the knots come to lie on the left side.

sucker can be placed through the tricuspid valve into the right atrium and steadied in such a manner that it not only keeps the field bloodless but also assists in exposure. Other retractors inside the heart are not usually utilized, but movable structures, particularly chordae tendineae, may be manipulated by gently grasping them with fine tissue forceps.

Once exposure has been obtained through the right ventriculotomy, the surgeon must orient the defect with relation to the *left* ventricular structures, since these are basic to the planning of the repair. First, the defect is identified as a high one by noting that it is immediately beneath the aortic cusps.

As mentioned previously, it is usually beneath the right and posterior cusps and their intervening commissure, but it may be beneath one of these more than the other. The correct identification of the position of the aortic ring is basic to the planning of the repair.

It is essential to realize that maximal protection to the integrity of the repair requires that closure be accomplished without undue tension. The power of the heartbeat can disrupt a repair done either with or without a prosthesis if the repair is made in an improper direction. These defects are usually ovoid, and the line of repair must be parallel to the long axis of the oval. This line

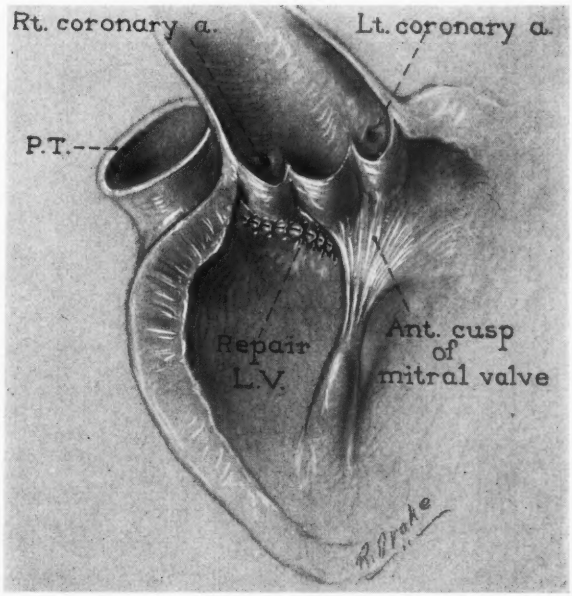

Fig. 3.—Completed repair of a high ventricular septal defect by the direct-suture technique, as seen from the left ventricular aspect. Note the relation of the direction of the resultant suture line to the aortic-valve ring.

is usually parallel to the aortic ring and transverse to the outflow tract of the left ventricle (Figs. 3 and 4). It is impossible to overemphasize the importance of this proper line of repair. To accomplish repair in this proper direction, the surgeon must constantly judge the left ventricular relations of the defect while working through the right ventriculotomy opening.

As the defect is viewed from the left ventricular aspect, its upper margin consists essentially of aortic ring and its lower margin of ventricular septum. When viewed through a right ventriculotomy incision, the previously described upper margin lies superiorly and to the patient's right, and the lower margin lies inferiorly and to the patient's left. When the ventricular septum is viewed from the left ventricular aspect, the central fibrous body lies in the region of the extreme posterior corner of the defect, or to the viewer's right. When

viewed from the right ventricular aspect, it is situated far posteriorly and beneath the aortic ring in a relatively inaccessible angle. This inaccessible angle is of critical importance in obtaining a complete and secure repair. It is essential that the repair begin in this region, for it is quite impossible to expose the posterior aspect of the defect properly and hence to accomplish complete repair if the suturing is begun at its opposite or anterior end. Furthermore, by beginning at this angle, this portion of the repair can actually be performed from the left ventricular side by working through the septal defect, thus obtaining not only additional space but also improved orientation which a left-sided approach provides.

Fig. 4.—Necropsy specimen of a high ventricular septal defect repaired at operation by the direct-suture technique, as viewed from the left ventricular aspect. The suture line is partially obscured by the overhanging aortic valve cusps. The patient died 48 hours after operation as the result of an associated severe congenital deformity of the mitral valve which could be only partially corrected. The defect was a very large one, but the repair is obviously without tension and structures are not distorted.

Only after study of the defect and its relations is completed should suturing begin. Number 3-0 silk threaded into a small sturdy needle (Adson dural needle) is employed. Direct suture is at present the technique chosen to repair nearly all of these high defects, including those which are very large. Since it is not certain that use of a prosthesis is to be completely abandoned, this technique of repair also is described.

Direct Suture.—The first stitch in a direct repair is begun by placing the tip of the needle holder through the ventricular septal defect into the left

ventricle, and then passing the needle from left to right through the ventricular septum at its junction with the central fibrous body and tricuspid ring. To complete this first stitch the needle is passed from right to left through the corresponding fibrous tissue superiorly at the junction of aortic-valve ring with tricuspid-valve ring. This suture thus grasps tissue at each extremity of the central fibrous body, and is tied with just sufficient tension to approximate the tissues snugly. This is the key suture—the principal one that assures a complete repair (Fig. 2, *a*).

Further interrupted sutures are then similarly placed at intervals of 2 to 3 mm. Each of these is first passed through the superior margin of the ventricular septum (or the lower margin of the defect) from the left ventricular aspect toward the right. Superiorly, each suture passes through the surprisingly tough tissue of the free margin of the aortic-valve ring, care being taken not to stitch into aortic-valve cusp (Fig. 2, *b*). Working in this fashion, each suture successively placed for repair of the defect lies more anteriorly than the previous one.

Because of the fact that each of the sutures placed thus far passes from the left ventricle to the right and then back again, the knots come to lie on the left ventricular aspect. This has not resulted in discernible undesirable sequelae.

After about half of the defect has been closed in the manner described, it will be found that the aortic ring gradually fuses with the deep surface of the muscle of the crista supraventricularis and disappears behind that structure. The crista itself at this point appears to form the upper margin of the defect, and there is no contraindication to regarding it as such. Therefore, a change is made in the method of suturing. The stitches now are made entirely from the right ventricular aspect, passing through the lower edge of the crista supraventricularis above and through the ventricular septal margin below, with the knots now lying on the right ventricular surface of the septum (Fig. 2 *c*). As these are placed, the remainder of the repair is progressively completed (Figs. 2, *d*, 3 and 4). As the final sutures are placed, it is important that the left ventricle be filled with blood and that residual air be expressed, it sometimes being necessary to inject blood into the left ventricle through the defect before the last sutures are tied.

Repair by Inserting a Prosthesis.—If the choice is made to utilize a prosthesis in the repair, the basic method is similar to that for direct repair. The prosthesis is first fashioned from a piece of polyvinyl sponge compressed to 3 mm. in thickness,* oval in shape, and only slightly smaller than the defect itself. The first suture is placed precisely at the same point as for direct repair, except that here, of course, each suture takes only one bite into tissue, passes through the polyvinyl sponge and then is tied. As this first suture, placed in the region of the central fibrous body, is tied, the prosthesis is lowered into position. It is helpful to conceive of this first suture, when tied, to be functioning as a hinge upon which the polyvinyl sponge can swing just as a

*Compressed to approximately a fourth of its original thickness.

trap door would swing on its hinge. Initially, this door is held open toward the right and against the tricuspid valve, thus leaving the ventricular septal defect open and permitting the placement of further sutures from the left ventricular aspect (Fig. 2, *a'*). Further simple interrupted sutures are thus placed both superiorly and inferiorly about the deep margins of the defect (Fig. 2, *b'*). Again, as the aortic ring is followed superiorly and anteriorly, it fuses with the musculature of the crista supraventricularis, and at this point the unattached portion of the prosthesis is swung to the left, completely closing the defect and permitting the remaining sutures to be placed and tied from the right ventricular aspect (Fig. 2, *c'*).

SUMMARY

High ventricular septal defects with little or no septal remnant beneath the aortic ring present technical difficulties in their complete repair. An accurate and reproducible technique is described whereby the complete correction of such defects can be accomplished.

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COARCTATION OF THE AORTA WITH A REPORT OF AN UNUSUAL OPERATIVE COMPLICATION

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THE majority of patients with coaretation of the aorta may be treated surgically with a good chance of obtaining a satisfactory result. Inasmuch as the average life expectancy of the untreated case is approximately 30 years, it behooves all physicians to endeavor to discover the patients with this defect early so that surgery can be performed before irreversible changes have occurred. The infantile type poses a more serious problem, since there is a long coaretate segment and, in addition, there are often associated cardiovascular defects. The surgical mortality rate is highest in the patients operated upon in infancy or early childhood and after late adolescence, or early adult life. When surgery is delayed until after age 30, a more serious appraisal should be made before the decision to proceed is made. The ideal age for surgical correction is perhaps in late childhood when the aorta has attained a substantial size and before serious degenerative changes have taken place. The over-all operative mortality rate at the present time is somewhat less than 10 per cent and may be expected to decrease significantly as earlier diagnosis and surgery is achieved.

The method of treatment of coaretation is entirely dependent upon the length of the narrowed segment, the availability of grafts, the position of the involved area, and the degree of associated cardiovascular defects of a congenital nature. The method proposed by Blalock and Park[1] involves an anastomosis between the left subclavian artery and the distal aorta. Crafoord and Gross,[2-4] in 1944, each operated upon patients by reseeting the coaretation and effecting an end-to-end anastomosis of the aorta. This method has continued to be the most effective method of surgical correction of coaretation and is used whenever possible. With the advent of common use of both homografts and synthetic plastic grafts, longer defects can now be bridged without great difficulty.

The majority of operative complications which result in death are those due to the direct effect upon the heart at the time of surgery. These include cardiac arrest, ventricular fibrillation, irreversible shock, and heart failure.

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Second in importance as a cause of death is disruption of the anastomosis, and third is operative hemorrhage which is usually initiated by trauma to the thin, friable, dilated intercostal vessels.

There is a miscellaneous category of operative complications which comprise a significant percentage, but are too varied in their nature to be covered in this discussion.[5-8] Among these complications is the entity known as necrotizing arteritis as described by Ring and Lewis.[9]

The case which is reported herein represents an example of a complication previously unlisted in the surgical literature to the best of the authors' knowledge. During surgery, the patient developed an extensive rupture of the proximal aortic arch with survival and satisfactory end-result.

CASE REPORT

R. C. W., a 21-year-old white man, was transferred to this hospital with a diagnosis of coaretation of the aorta. Several months prior to admission and just prior to discharge from the army, a physical examination revealed a blood pressure of 140/110 mm. Hg in both upper extremities. Presumably the patient was normotensive on previous examinations during and prior to military service. The patient had no significant complaints other than occasional numbness or a tingling sensation in the lower extremities for approximately two months. He stated that his feet had always been cold but he denied any weakness and had been active in athletics during his preparatory school days. On physical examination, the pressure in the right arm was 150/105 mm. Hg and that of the left arm 145/105 mm. Hg. Pressures in the legs were unobtainable. The body was muscular and well proportioned with no evidence of lower extremity muscular atrophy. The peripheral pulses of the lower extremities were not obtainable. The neck veins were not distended. There was no ankle edema. The chest was clear. There was no significant cardiomegaly but a harsh Grade 3 systolic murmur was noted over the aortic area. The skin of the lower extremities was damp, moist, and cold.

Laboratory data: Chest x-ray revealed enlargement of the cardiac silhouette. There was some rotation of the heart with recession of the aortic knob. There was minimal notching of the ribs in the midthorax. An esophagram revealed no fluoroscopic evidence of displacement or other significant abnormality. An electrocardiogram was considered to be within normal limits.

Shortly after admission, an angiocardiogram was performed by rapidly injecting 50 c.c. of 90 per cent Hypaque through a No. 14 polythene catheter with its tip in the axillary vein. The film taken 7 seconds after this injection shows a typical segment of coarctation immediately beyond the take-off of the left subclavian artery (Fig. 1). Several large collateral vessels were also visualized.

Following adequate preoperative preparation, the patient was taken to surgery and in the right lateral position a left infrascapular incision was made. The dissection proceeded down to the pleural cavity which was entered without difficulty. Numerous large vessels were encountered during this procedure, all of which were securely ligated. On inspection of the great vessels, the arch of the aorta was found to be somewhat smaller than normal and the left subclavian artery was enlarged to approximately the size of the aortic arch. There was a short complete coarctation 3 cm. below the take-off of the left subclavian artery. The third intercostal arteries were small, but the fourth intercostal arteries and the internal mammary arteries were markedly dilated. The mediastinal pleura was incised over the aorta extending upward between the vagus nerves and left subclavian artery. Mobilization of the aorta and the left subclavian artery was being accomplished when a cardiac arrest was noted.

The lung was retracted from the heart surface and cardiac massage was immediately begun. However, it was noted that pressure against the myocardium was very resistant. In order to secure more effective massage, it was decided to incise the pericardium and upon so doing a large quantity of blood gushed from the pericardial cavity. Blood continued

to exude in large volume and consequently the pericardium was extensively incised and explored. A long rent, approximately 4 cm. in length, was identified on the right posterolateral aspect of the ascending aorta with its lowermost extension about 2 cm. superior to the aortic valve. The position of the rent made control of the hemorrhage most difficult. A running suture of No. 1 silk was used to close the defect and, following this, several points of leakage were reinforced with No. 4-0 arterial silk. There was moderate amount of ooze from the sutured defect which gradually ceased upon pressure over the area. Immediately after incision of the pericardium and release of the acute cardiac tamponade the heart regained its normal rhythm and this continued throughout the period of observation. Upon

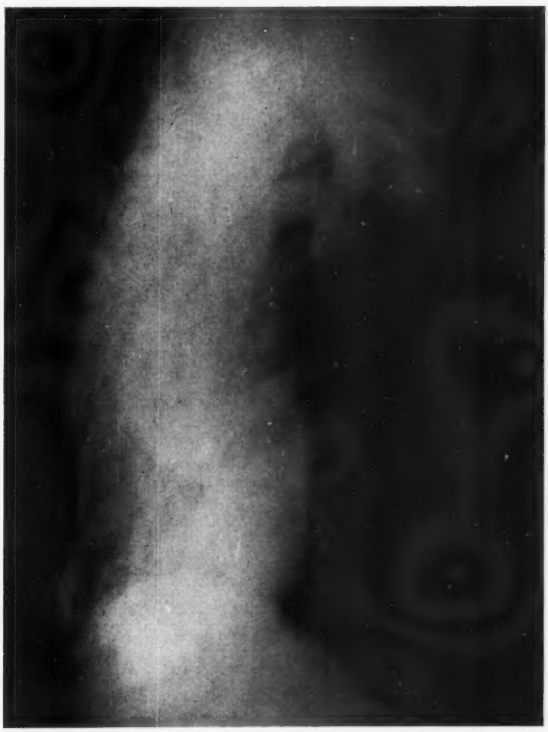

Fig. 1.—Angiocardiogram showing complete coarctation and large left subclavian artery.

closure of the rupture and coincident with rapid transfusion of 2,500 c.c. of whole blood, the blood pressure again returned to normal where it remained throughout the completion of the procedure. In spite of the grave situation which existed, it was felt that resection of the coarctation would lessen considerably the pressure upon the sutured aortic defect. Consequently, the subclavian artery and aortic arch were mobilized, the ligamentum arteriosum ligated, and the narrowed segment was excised. An end-to-end approximation was accomplished with posterior and anterior everting, running mattress sutures, reinforced at various points with interrupted mattress sutures. The clamps were slowly released following suture and the patient's condition remained satisfactory throughout the remainder of the closure. The patient received a total of 16 units of blood (8,000 c.c.) during and immediately after operation. Examination of the specimen revealed the aorta to be totally occluded (Fig. 2). Postoperatively, the course was characterized by moderate hyperpyrexia, blood pressure fluctuations, and tachycardia. In addition, there was an obvious clouding of the sensorium.

However, one week following the operative procedure, the patient appeared in all respects normal with the exception of episodes of tachycardia. The patient's memory returned without defect and his intelligence seems in no way disturbed. Tests by psychologists rate him as a high normal. The use of heavy dosage of antibiotics was continued for 3 weeks following surgery. After only minimal difficulty with postoperative hypotension the blood pressure has stabilized at about 105/70 mm. Hg, and has remained at this level for 3 months postoperatively.

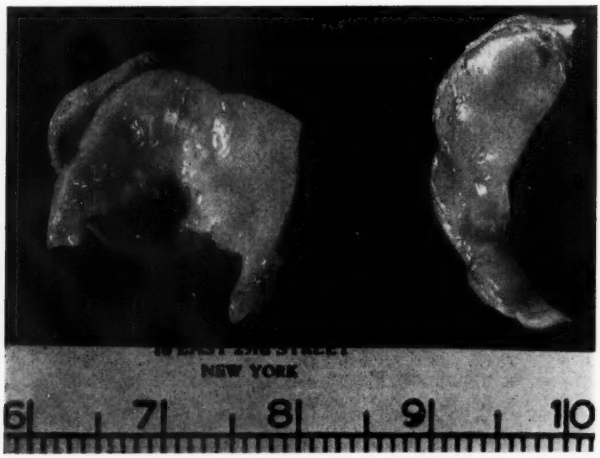

Fig. 2.—Excised coarctate segment.

DISCUSSION

It has been reported by Reifenstein and co-workers[10] that, of 104 autopsied cases of adult type of coarctation, approximately one fourth of patients died from rupture of the aorta. An additional one fourth succumbed from endarteritis or endocarditis, one fourth from cerebral vascular accidents, and one fourth from causes not related to the cardiovascular system. Upon reconsideration of the events immediately prior to operation, it was recalled that the patient complained of mild to moderate substernal pain somewhat to the left of the mid-sternal line. This had been noted on several occasions for approximately one week prior to operation, and it was felt by the authors that this represented apprehension which the patient was experiencing as the date of his surgery approached. In retrospect, it appears that the pain indicated the first events of what was to be a rupture of the aorta. Due to slight blood pressure increase during the minimal trauma associated with mobilization of the aortic arch, the rupture did occur and fortunately was quickly identified and repaired. The anesthetist reported no significant fluctuation in blood pressure prior to the cardiac arrest, although, presumably this could have been momentary and not noted during the routine blood pressure determination. Although further examination during the procedure was limited because of the gravity of the situation, there were no other outstanding cardiovascular defects noted.

SUMMARY

A brief discussion of the necessity for surgical treatment for coaretation of the aorta is given. A most unusual and previously unreported complication occurred at operation, namely, rupture of the proximal aortic arch. This was successfully sutured and a satisfactory result obtained.

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RECONSTRUCTION OF THE TRACHEA AND ITS BIFURCATION

An Experimental Study

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LARGE tracheal defects necessitating circumferential reconstruction present a difficult problem, especially when the carina is involved. They must be repaired with some material which is airtight and semirigid and with a minimum of tissue reaction. Daniel[6] has shown that tubes of Vitallium, stainless steel, and glass will provide a temporary airway but will not give a good long-term result. Rob and Bateman,[9] Bucher and co-workers,[7] Edgerton,[10] and Keshishian and co-workers[5] have used stainless steel and tantalum mesh, while Belsey[8] used a wire coil wrapped with host tissue for tracheal replacement. Excellent experimental results have been obtained by Ekeström,[2] who embedded subcutaneously a plastic frame, placed over a plastic tube wrapped in fascia, in the experimental animal for 3 to 12 weeks prior to the reconstruction. At the reconstruction, the inner tube was removed when an autologous connective tissue tube, reinforced with a plastic (Teflon) frame, was obtained.

EXPERIMENTAL RESULTS

1. *Homografts.*—Preserved homografts have been used earlier by Keshishian, Blades, and Beattie.[5] These tracheal grafts were preserved in modified Tyrode's solution with antibiotics and maintained at –4° C. No grafts were older than 14 days. These homografts seemed to be well healed after one week but, in the period of 3 weeks, there was a beginning liquefaction and necrosis, leading to stenosis and death. We have tried to desensitize homologous transplants of trachea, in the following manner.

Segments containing 10 to 11 rings of cartilage of the trachea were freeze dried and then implanted in the trachea in 4 dogs. All dogs died due to degenerative changes of the homografts. The rings of cartilage rolled into the lumen of the trachea, causing stenosis and death (Fig. 1). In 2 dogs, a formalin-fixed tracheal homograft, either 40 or 4 per cent formalin, was used with the same disappointing result. In another 2 dogs, a tracheal homograft preserved in 95 per cent alcohol was used, with liquefaction and necrosis developing in 2 to 4 weeks.

2. *Autografts.*—In a 15 Kg. dog, a piece of trachea containing 13 rings was resected and then a right pneumonectomy was performed. The right main bronchus was then used for the reconstruction of trachea in the following manner. The autograft was first sutured to the transected end of the trachea.

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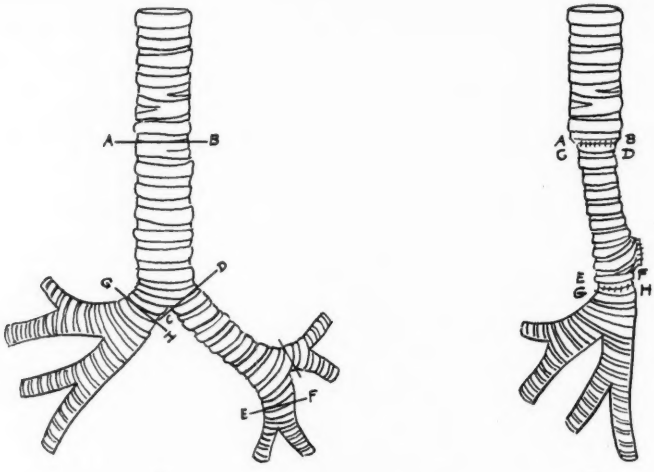
The distal part of the autograft was then sutured to the left main bronchus. Necrosis occurred in that autograft. In another dog, a left pneumonectomy was performed first and the left main bronchus was then used as an autograft replacing 8 rings of the trachea (Fig. 2). In this case, first a left thoracotomy was performed for the pneumonectomy, then a right thoracotomy for the tracheal replacement. Necrosis occurred, however, in the autograft (Fig. 3, *A* and *B*). In the third dog, first a left thoracotomy was performed with extirpation of the left upper lobe and with a direct anastomosis of the left upper lobe bronchus

Fig. 1.—Specimen from dog, in which a freeze-dried tracheal homograft was inserted after resection of 10 rings of cartilage of the trachea. After 15 days, necrosis of the mucosa occurred and the cartilage rings were lying loose inside the lumen of the trachea, causing obstruction and stenosis breathing. Fifteen days after the insertion, the other specimens had a similar appearance.

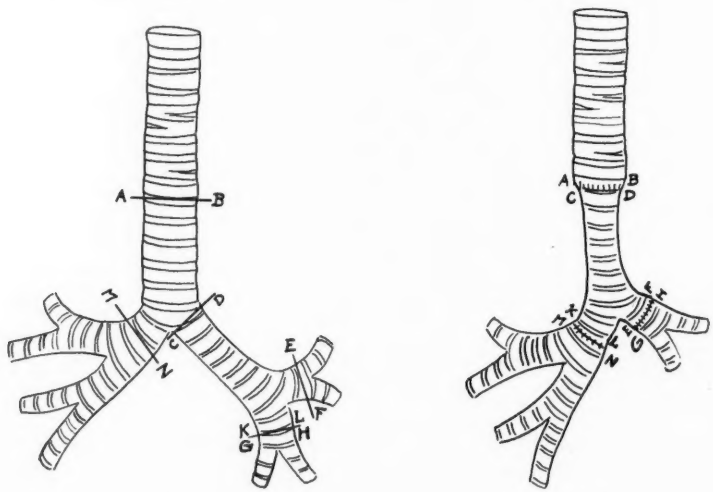
to the left lower lobe bronchus. Thus, the lower lobe remained in the left thorax. The main bronchus was divided at the level of carina. Then a right thoracotomy was performed, the trachea resected, and the left main bronchus used for tracheal reconstruction. The divided bronchus to the left lower lobe was then anastomosed to the right lung. Thus, a complete reconstruction of the tracheal bifurcation was obtained. A necrosis occurred, however. As necrosis occurred in a main bronchus without connection to its lung when it was used for reconstruction of the trachea, this necrosis was thought to be due to a lack of blood supply through that pulmonary artery and the experiments were changed in the following way.



A. *B.*

Fig. 2.—*A*, The left main bronchus was used as an autograft for replacement of trachea. First, the left thoracotomy with left pneumonectomy was performed. The left main bronchus was then, by means of a right thoracotomy, used for replacement of the trachea.

B, The left main bronchus has been inserted in place, connecting the distal end of trachea to the right main bronchus.

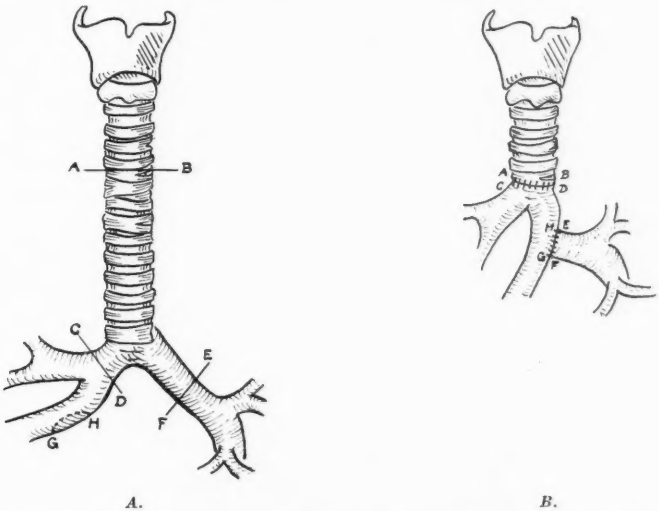


A. *B.*

Fig. 3.—*A*, First, a left thoracotomy was performed with resection of the left upper lobe. The left upper lobe bronchus was then anastomosed to the left lower lobe bronchus. The left main bronchus was divided at the level of the carina and the left thoracotomy wound was closed.

B, A right thoracotomy was then performed. The left main bronchus was drawn up in the wound and anastomosed to the distal cut end of the trachea. The original left lower lobe bronchus was then anastomosed to the right main bronchus. Thus, a complete reconstruction of trachea and carina, with first parts of both main bronchi, was obtained. Necrosis occurred in the autograft.

3. *Direct Anastomosis.*—Through a right thoracotomy, a wide dissection was made of the trachea, the carina, and both main bronchi. After resection of 12 tracheal rings, the right main bronchus was sutured end-to-end to the trachea. Then the left main bronchus was sutured end-to-side to the intermediate bronchus below the origin of the right upper lobe (Fig. 4). Thus, both lungs were preserved yet the lower part of trachea with the carina could be reseeted. This proved to be a rather difficult procedure in a small dog as the quality of the left main bronchus was not good enough for anastomosis end-to-side to the right intermediate bronchus. The cartilages of the left main bronchus were too soft and too small. The experiment was therefore repeated in a 30 Kg. dog which made an uneventful recovery for 3 months and then died due to infection and atelectasis. There was an abscess around the suture line to the left lung with fibrosis and degeneration in the main bronchi. This, however, seems



A. *B.*

Fig. 4.—*A*, The trachea, carina, and first parts of the left and right main bronchi were resected. Then, an end-to-end anastomosis was made between the distal part of the trachea and the right main bronchus. Then, the left main bronchus was anastomosed end-to-side to the intermediate portion of the right main bronchus.

B, An anastomosis completed after resection of the trachea and carina with preservation of both lungs.

to be the most promising method in the resection of the carina with the distal part of the trachea, i.e., to do an end-to-end anastomosis between the right main bronchus and lower end of the trachea and then to implant the left main bronchus in the right main bronchus. Due to the good results obtained in 12 clinical cases of bronchotracheal anastomosis, this method of direct anastomosis for carinal reconstruction is considered the best method available. It will, however, be difficult to perform this operation in dogs due to the soft cartilage of the left main bronchus. A similar method has already been successfully applied to a clinical case by Mr. R. Barclay.

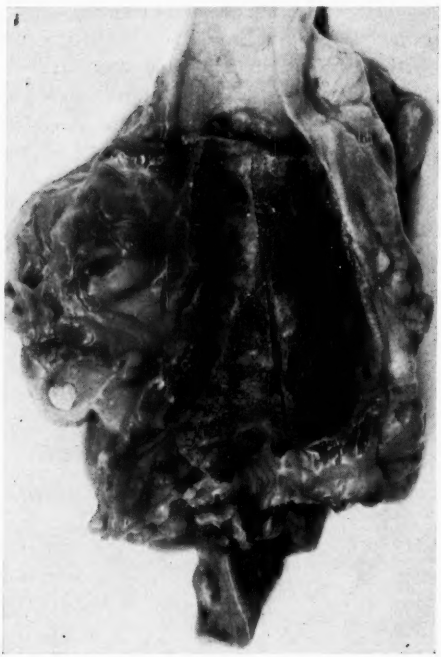


Fig. 5.—Specimen from a bifurcation prosthesis of reinforced Ivalon sponge lying loose in an abscess formation.

Fig. 6.—Another bifurcation prosthesis which, after 10 months, caused death. It was lying loose in an abscess having perforated the esophagus.

Juvenelle has already shown, in 1951, that it is possible to remove the lowermost part of trachea with its bifurcation. But with a resuture of both main bronchi end-to-side to the trachea of a dog, it is not possible to mobilize the left main bronchus for any significant distance. It is, however, possible to remove a longer section of trachea when an end-to-end anastomosis between right main bronchus and the trachea is performed and the left main bronchus is sutured into the right main bronchus, end-to-side. The suture line must not be under too much tension, otherwise it may cause a stricture to form. From these dog experiments, it seems that there is a more pronounced tendency for stricture formation when an end-to-end anastomosis of a main bronchus to the trachea is made under tension as compared to a bronchotracheal anastomosis.

Fig. 7.—Perforation of the abscess, around a prosthesis, into the esophagus. In another case, there was erosion of the carotid artery.

In 1950, Ferguson and associates[1] reported a study of primary tracheal anastomosis. They concluded that when stenosis, following repair, did not exceed one third of the original lumen, there were no distressing symptoms. If, however, the stenosis occluded the trachea to one fourth of its normal circumference stridor and respiratory infection would follow.

4. *Plastic Prosthesis.*—In a series of dogs, a plastic tube prosthesis of Ivalon sponge made over of a frame of a stainless steel spiral was used for tracheal reconstruction. This prosthesis was made in the following way:

First, a layer of Ivalon sponge was rolled over glass or plastic tubing. Then a coil of stainless steel was rolled over the Ivalon. Over this, another layer of Ivalon was rolled. Then, this tube of Ivalon was compressed by gauze and autoclaved. In this way, grafts of the tracheal bifurcation could easily be constructed with the desired angle between the 2 main bronchi. The primary result

with this prosthesis was excellent. However, there was no long-term survival, although the longest survival was 12 months. After a period of 6 to 12 months, this plastic prosthesis either caused a pressure necrosis with perforation of a big artery or eosophagus, or there was an inflammation around the prosthesis so that the prosthesis was actually "floating" in an abscess (Figs. 5, 6, 7). Although some of these dogs seemed in excellent condition up to one year, they then died either of hemoptysis, due to perforation, or infectious complications. Reconstruction of the tracheal bifurcation seems to be especially difficult as it is difficult to obtain the exact angle of the main bronchi; if this angle is not ideal, pressure necrosis will occur at the distal suture line.

DISCUSSION

Reconstruction of the carina with plastic tubes of Ivalon, reinforced with a stainless steel coil, has given a very disappointing result. Although the dogs recovered from the operation and seemed to go through an uneventful recovery for some months, all died within a year. The cause of death was pressure necrosis of arteries or esophagus, or late infections. It was especially difficult to obtain a correct angle between the main bronchi to prevent the distal suture line from pressure necrosis. Therefore, homotransplants were tried but they became necrotic. An autograft of a main bronchus also necrotized when used for tracheal reconstruction. With the very good clinical experience we have from bronchotracheal anastomosis, it is evident that a main bronchus connected with its lung and receiving its blood supply through the pulmonary artery may be used for tracheal reconstruction. In dogs it is easy to resect the lower 12 tracheal rings and do an end-to-end anastomosis of the right main bronchus and trachea. If too great a tension is exerted on the suture line, a stricture formation at the anastomosis will result. In one clinical case of right-sided bronchogenic carcinoma, the left main bronchus was anastomosed to the trachea and after that a right pneumonectomy was performed which included the lower part of trachea and the first part of the left main bronchus. Considerable tension on the suture line resulted as the aorta prevented mobilization of the left main bronchus and a stricture developed. The right main bronchus can be mobilized higher without undue tension on the suture line. All anastomoses were made using 3-0 chromic catgut on atraumatic needles. In an earlier work on the bronchotracheal anastomosis, we found that silk sutures around an anastomosis caused suture granulomas with stricture formation. Catgut will diminish the incidence of suture granulomas considerably and is therefore routinely used for bronchial closure and anastomosis.

SUMMARY

An experimental study of the reconstruction of trachea and its bifurcation is reported. The use of Ivalon prostheses showed good early results but within one year all dogs died due to complications, such as pressure necrosis or infection. A direct anastomosis of the right main bronchus to the trachea, end-to-end, followed by an end-to-side anastomosis of the left main bronchus to the

right main bronchus is suggested as the best method to use whenever possible. The left main bronchus of the dog is, however, not suitable for an end-to-side anastomosis.

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BRONCHOGENIC CYSTS AND THE THEORY OF INTRALOBAR SEQUESTRATION: NEW EMBRYOLOGIC DATA

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SOLITARY or multilocular cysts of the lungs, filled with air or watery fluid and lined with bronchial epithelium, have long been recognized as congenital malformations because, on rare occasions, they have been found in premature and newborn infants. But, so far as the writer is aware, they have never been identified in embryos or in early fetal stages. The problem is further complicated by the frequent presence of infection and in the lower lobes, by the coincidental occurrence of a systemic pulmonary artery. For these reasons the debate still continues as to whether such cysts represent early developmental anomalies or must be attributed to environmental changes of the later fetal and postnatal periods.

For those who hold the former view the most popular theory (Ravitch[17]) seems to be that developed by Pryce and his associates,[15, 16] namely, that cysts represent "sequestered" masses within the lung, comparable to "sequestered" masses of pulmonary tissue found outside the lung. Both are conceived as arising from "traction on the capillary nets of the developing (bronchial) tree by aberrant arteries that anchor the mass while the lung is undergoing developmental shifts." Certainly, the relative frequency with which cysts and accessory pulmonary arteries occur together in patients requiring pulmonary excisions has to be explained; but it does not follow that the two must stand in the relation of cause and effect—especially in view of the fact that they may occur separately. Nor can cysts of the upper lobes be explained by this hypothesis. Nevertheless, all who have wrestled with this problem are indebted to Dr. Pryce and his associates for having recognized the relatively frequent association of these two anomalies in patients and for having presented a theory that has stimulated so much discussion.

The two principal objections to the theory are (1) that it is not consistent with what is known about developmental processes in human and other mammalian embryos (Boyden[1]), and (2) that intra- and extralobar sequestration (lower accessory lung) differ from each other in at least seven ways and therefore "are not related by any mechanism which involves traction by the systemic aberrant artery" (R. Abbey Smith[18]).

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Obviously, the only hope of reaching an understanding of these lesions is to "catch" such anomalies in the process of formation. With this in view the writer examined part of the large collection of sectioned human embryos in the Carnegie Laboratory of Embryology in Baltimore.* By great good fortune, cysts were found in the upper lobes of a 31 mm. human embryo, and an accessory pulmonary artery in a slightly older 41 mm. fetus.

LUNG CYSTS IN AN EMBRYO OF HORIZON XXIII

The 31 mm. male embryo shown in Fig. 1 is a perfect specimen obtained at operation for tubal pregnancy on a 31-year-old Negro woman. The recorded menstrual age of the embryo was 9 weeks, and the ovulatory age of this horizon was 46 to 48 days (Streeter[20c]). The only malformation observed by the writer in this embryo was a pair of lung cysts.

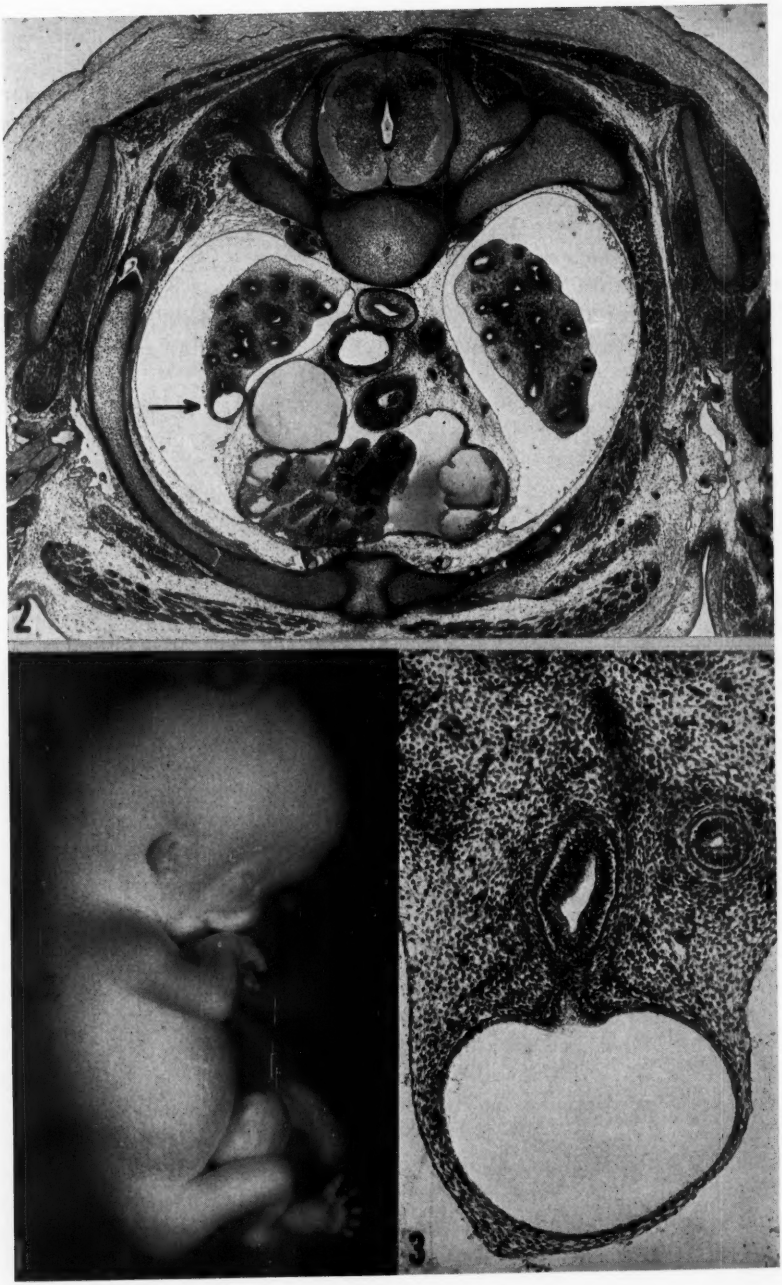
Fig. 2 is a transverse section through the thorax in the region of the superior mediastinum. The lungs are in the so-called glandular period of development, characterized by early dichotomous budding of the bronchial trees. Branches are lined solely by pseudocolumnar epithelium having several layers of nuclei. At this stage the lungs do not fill the pleural cavities and lie mostly behind the heart.

A single cyst was present on the ventral (anterior) margin of each upper lobe. These were symmetrically placed on right and left lobes and represent an enlargement of a terminal bud of the anterior segmental bronchus. The right one (arrow, Fig. 2) runs through 34 sections 10μ thick (thought to be nearer 12μ) and therefore measures approximately 0.4 mm. in diameter. The patent stalk to which it is appended runs through only 6 sections.

The cyst of the left upper lobe is shown in Fig. 3, enlarged 100 times. It runs through 42 sections and measures approximately 0.5 mm. in diameter. The stalk to which it is attached runs through only 4 sections. If the stalk of the left cyst is patent, it is just barely so. (The section photographed passes to one side of the lumen.) The photographs reveal the fact that both cysts are under fluid pressure; for, proximally, the epithelium of each is cuboidal, and distally, it is flattened.

From a mechanical standpoint the explanation might be sought in the narrowing of the necks of the terminal bronchial buds, in accordance with the Laplace law of pressures in cylindrical tubes (Burton[4]). But, strangely enough, this vase-shaped form of the growing bronchial buds is the prevailing pattern (Fig. 4). Also, these cysts have developed before any smooth muscle fibers have formed around the bronchi. Nor can one attribute the formation of these cysts to any ischemic condition, for the bronchial epithelium is surrounded by an abundant plexus of capillaries (see Fig. 3). Aside from these categories it is difficult to find an explanation. Naturally one asks why cysts do not appear in other portions of these lungs. This raises the question of time specificity in teratogenesis (Wilson[24]). Since there are critical periods in

*For this privilege the writer is much indebted to Dr. James D. Ebert, Director of the Laboratory.



Figs. 1-3.—*For legends see opposite page.*

development when certain agents can interfere with development, it may be conjectured that at a time when these particular buds of the anterior segment were growing fastest (B^2b, Fig. 4) something interfered with the metabolism of the embryo. Once an accumulation of fluid started, the narrow neck could act as a ball valve, ballooning up the terminal segment. In a tubal pregnancy, such as this, the ectopic position of the chorionic vesicle might be a determining factor; but similar interference could arise in normally implanted embryos, witness the experimental production of agenesis of the lung in fetuses of rats whose mothers have been deprived of vitamin A. (See discussion in article by Thomas and Boyden[21]).

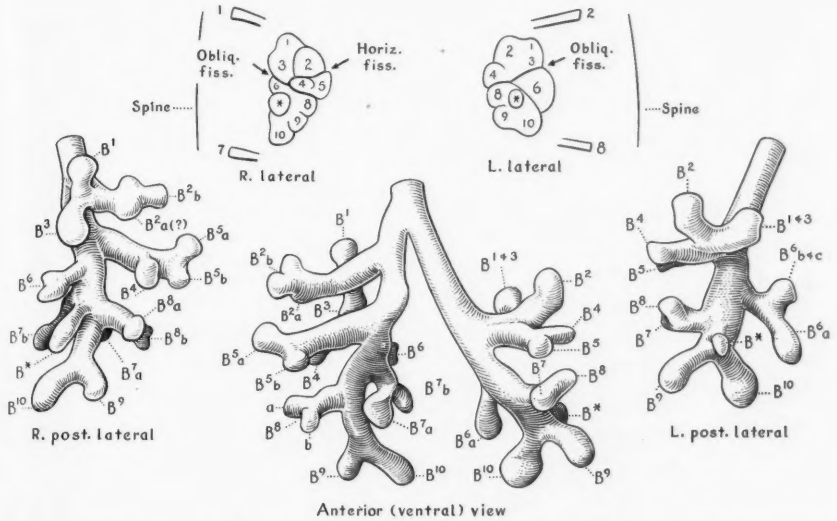


Fig. 4.—Wax model of the bronchial tree of a 13 mm. human embryo of Horizon XVII. (After Fig. 108 of Boyden's Segmental Anatomy of the Lungs, Blakiston Div., McGraw-Hill Book Co., 1955) B^1 to B^{10}, developing segmental bronchi.

Be that as it may, these cysts could well have developed into solitary bullae which years later could have ruptured spontaneously. If more buds had been involved, these in turn could have compressed adjacent segments and thus have prevented the formation of alveoli. Eventually the whole lobe might have become a mass of cysts. (See cases of balloon cysts of infants

Fig. 1.—Photograph of a 31 mm. human embryo (Carnegie Coll., No. 9226) which contained a symmetrical pair of lung cysts (×3).

Fig. 2.—Transverse section through the superior mediastinum of embryo No. 9226 (×15). The arrow points to a cyst on the anterior margin of the right upper lobe. This is a distended terminal bronchial bud of the anterior segment. Since the left lung is normally smaller than the right, the corresponding cyst on the left side appeared at a slightly lower level (see Fig. 3).

Fig. 3.—The corresponding cyst on the anterior margin of the left upper lobe (×100). The section passes slightly to one side of the lumen connecting the distended terminal bud with its parent bronchus.

occupying the entire middle lobe, or the entire right lower lobe, or just the superior segment with compression of the rest of the right lower lobe (Potts and Riker,[14] Burnett and Caswell,[5] and others too numerous to mention).

Incidentally, it should be noted that no accessory pulmonary artery was present in this embryo, nor in the infant cases just mentioned. This brings us to description of the first case of a systemic pulmonary artery observed in early fetal stages.

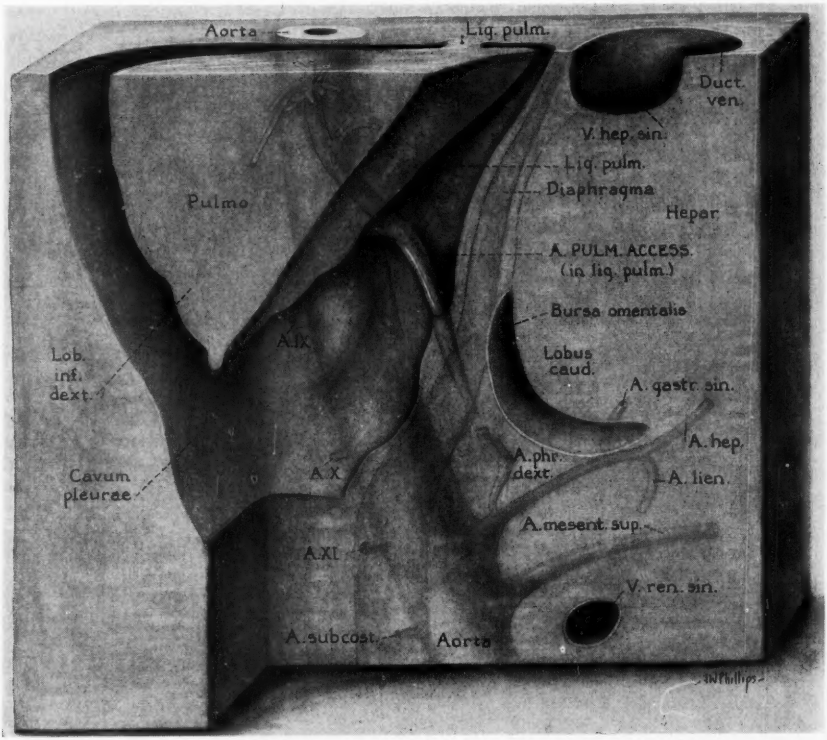


Fig. 5.—Graphic sagittal reconstruction of a 41 mm. human embryo (Carnegie Coll., No. 6361 [×30]) showing the topographic relations of an accessory pulmonary artery of aortic origin. The principal plane of section is just to the right of the aorta. On the viewer's left, the plane of section lies parallel to the first plane but cuts through the middle of the right lung.

THE ACCESSORY PULMONARY ARTERY

A systemic pulmonary artery arising from the aorta at the level of the tenth intercostal artery, and supplying the peripheral bronchi of the right posterior basal segment, was found in a young fetus of the Carnegie Collection having a C.R. length of 41 mm. It is of interest to anatomists that this was obtained by Dr. Lawrence Wharton at the hysterectomy of a 32-year-old white woman, and that the embryo was dissected out personally by Dr. Streeter to whom we are indebted for so many *Contributions to Embryology*

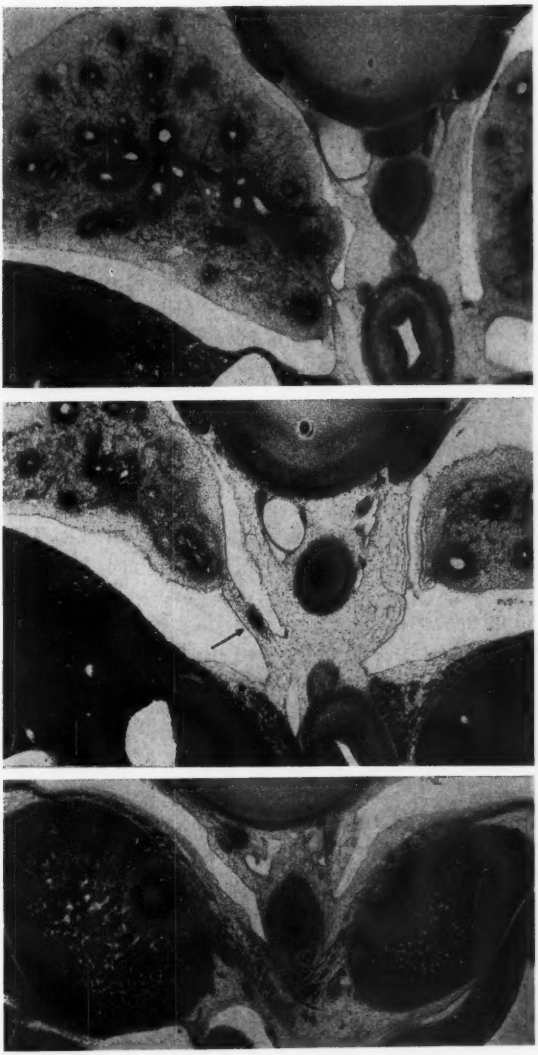


Fig. 6.—Transverse section through the right lung of Embryo No. 6361 at the level where the accessory artery bends downward (cf. top level of Fig. 5). The systemic pulmonary artery is indicated by an arrow (×22; reduced ½).

Fig. 7.—Transverse section of Embryo No. 6361 showing accessory artery (see arrow) ascending to the lung through the pulmonary ligament (×22; reduced ½).

Fig. 8.—Transverse section of Embryo No. 6361 showing accessory artery (see arrow) coursing between the leaves of the crus of the diaphragm.

The patient had had one miscarriage 8 years previously, and 2 normal births at term. Implantation was on the anterior wall of the uterus, and there was "a slight amount of blood in the decidual cavity."

The portion of the embryo containing the artery and adnexa was reconstructed graphically by the author and rendered in three dimensions by Miss Jessie Phillips. The 3 mm. block shown in Fig. 5 represents 60 transverse sections of 50μ each. After the artery leaves the ventral side of the aorta it courses ventralward through the leaves of the diaphragm (see Fig. 8, arrow). (Presumably, therefore, it would have emerged from the diaphragm if the embryo had reached maturity.) Then it enters the free margin of the right pulmonary ligament, from which it bulges (see level of guide line labeled, *A. PULM. ACCESS.*, Fig. 5). Just above this level it is concealed within the broader portion of the pulmonary ligament (Fig. 7). Shortly afterward it leaves the ligament to invade the right lower lobe of the lung. At the height indicated in Figs. 5 and 6 it sprays out and showers down like a Roman candle to supply the lower branches of the posterior basal segment—specifically $B^{10}a$ and $B^{10}b$ (Boyden[2]). At the same level (Fig. 9), it sends smaller branches upward to supply upper rami of B^{10}.

Fig. 9 represents a graphic reconstruction of all pulmonary arteries of the right lung below the level of the upper lobe bronchus. Only the arteries of the accessory subsuperior bronchus *BX* (10)*,[19] and those of the aortic pulmonary artery have been drawn in their entirety—that is, so far as they could be traced.

Five principal observations may be made. First, the peripheral portions of both pulmonary arteries are in process of formation. They can be traced only so far as smooth muscle has formed around them. Beyond this point—which is well behind the terminal bronchial buds—they are lost in a minute capillary or (arteriolar) plexus. That is why the specific segmental arteries of an embryo cannot be identified as such, much before the stage of Horizon XIX (Wells and Boyden,[23] Plate 4 and text).

Second, the branches of the aortic pulmonary artery resemble closely those of the right pulmonary artery—both in their structure and in their spatial relations to the bronchi they supply—differing importantly only in origin and in course, but not in structure.

Third, the unbranched portion of the posterior basal bronchus is unaccompanied by any artery. In other words, the posterior basal artery is absent as such except for the small artery, *AX* (10)*, which supplies the accessory subsuperior bronchus. The aortic pulmonary, which is taking its place, does not extend beyond the uppermost budding branches of the posterior basal bronchus. There is thus a "no man's land" or interarterial zone which

Fig. 9.—Graphic reconstruction of the whole pulmonary arterial system of the right lung below the level of the upper lobe bronchus (×30; reduced ½). The upper half of the figure shows the definitive right pulmonary artery. Only the arterial rami accompanying the accessory subsuperior bronchus, *AX* (10)*, have been carried as far as they can be traced. The other segmental arteries are shown as cut off at a higher level. Normally, *AX* (10)* is a branch of the posterior basal segmental artery (A^{10}) but, in this embryo, A^{10} is a branch of the aortic pulmonary artery. The lower half of the figure shows the course and rami of this accessory pulmonary artery. *T III – T X*, levels of thoracic vertebrae, between the nuclei pulposi.

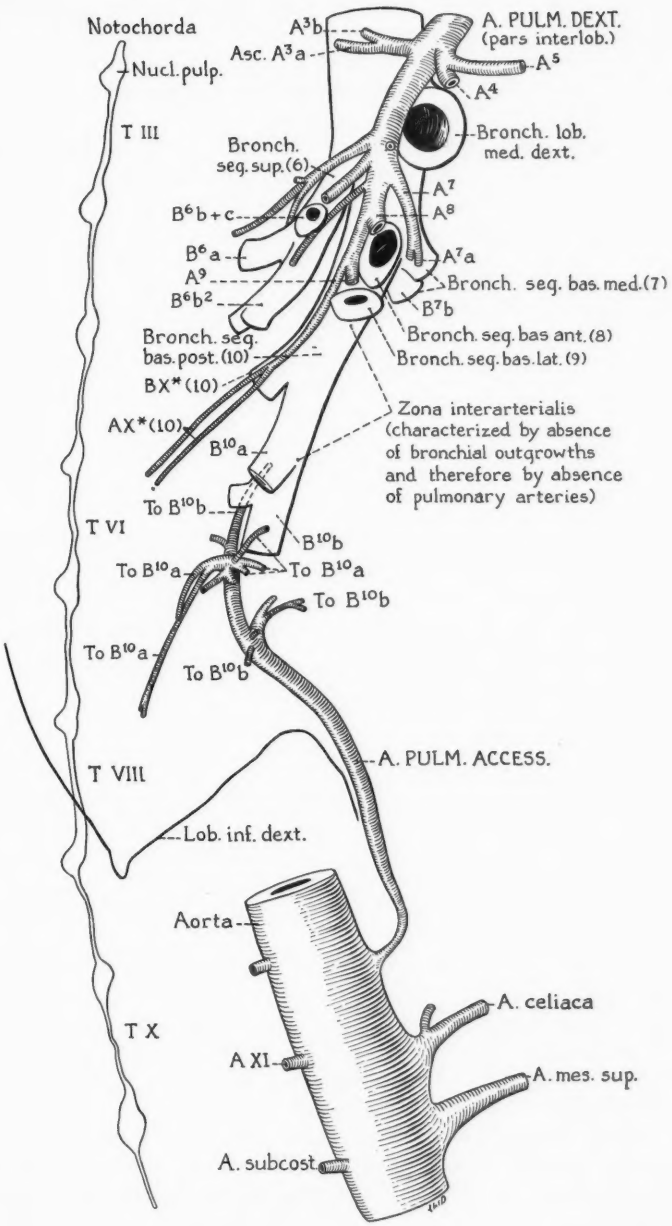


Fig. 9.—*For legend see opposite page.*

is devoid of arteries. To be sure, the blastema of this segmental bronchus contains minute capillaries which, presumably, will be connected with bronchial arteries; but these are not to be confused with the differentiating strands of the pulmonary arteries which take up a position that is posterolateral to the respective bronchi and lie well outside the blastema.

Fourth, there is no evidence of any extraneous factor, such as compression or atrophy, to account for the absence of the definitive posterior basal branch (A^{10}) of the right pulmonary artery. The explanation would seem to be a much simpler one—namely, that since the growing tips of pulmonary arteries terminate in the peripheral vascular plexus of the lung, the original area of distribution of the right pulmonary and the accessory artery has been moved apart by interstitial growth of the trunk of the posterior basal bronchus. (For elaboration of this idea, see "Discussion.")

Fifth, the most striking feature of the accessory artery is its relatively great length and its nearly vertical course. This, however, is only a temporary situation. Had the fetus lived, and had the lung grown down to its definitive position, the artery would have become nearly horizontal and would have resembled both in position and distribution that of Kinsella's case (see Boyden,[2] Figs. 120-122).

One may now ask, what was the original position of this systemic pulmonary artery. In the 4 to 5 mm. stage when it first appeared (age 28 to 30 days, Horizons XIII-XIV) it must have arisen at the level of one of the upper cervical vertebrae and have had the appearance of a more or less horizontal branch passing from the ventral side of the left aorta to the closely adjacent visceral plexus surrounding both the esophagus and trachea and the two lung buds.* Tobin has drawn such a hypothetical branch in his discussion of the origin of bronchial arteries (Tobin,[22] Fig. 1). The stage in question is that in which the lung buds and esophagus are closest together. At any later time it would be impossible for a ventral branch of the aorta to reach the bottom of the lung without traveling a most circuitous course. At this early stage, the celiac artery lies at about the level of the seventh cervical vertebra (Evans,[8] p. 648). Between this stage and the stage of the 41 mm. embryo (see Fig. 5), the accessory pulmonary artery and the visceral arteries below it will shift their points of origin along the ventral side of aorta faster than the lung grows downward (for discussion of this mechanism see Evans,[8] p. 648).

In summary, the following sequence of events can be established: first, the accessory pulmonary artery is approximately horizontal, lying in the region of the upper cervical vertebrae; second, its aortic end shifts with the visceral arteries, down along the ventral side of the aorta, until the artery becomes nearly vertical; third, the lung catches up with it and the artery to the base becomes nearly horizontal again. This shift in position explains why the upper portion of the accessory artery, in the stage shown in Fig. 5, meets the posterior segmental bronchus at an angle and then curves downward to supply its branches.†

*Cf. Congdon[7]; Buell[3]; Streeter[20a]; and Neill[12].

†A similar relationship of systemic arteries to bronchi has been reported recently by Halasz, Halloran, and Liebow.[9] They encountered six accessory pulmonary arteries in th

(Cont'd on opposite page.)

DISCUSSION

The sequence of events described above affords a general explanation of why Pryce's theory of the "capture" of bronchial buds by "anchored" arteries seems not to fit the facts of development. If traction on the bulbous tips of the posterior basal bronchus had occurred it should have been revealed at an earlier period; for at this stage of development, in this embryo, it is too late. Yet it has not occurred and the reason is obvious. The nature of embryonic shifts is such that no traction is exerted, even though an artery in the course of development meets a given bronchus at an angle (see Fig. 5). Shifts represent differential rates of growth with corresponding adjustments in the position of vascular trunks. If it were not so, no embryo would ever reach maturity.

Let us now examine the details of Pryce's theory, so far as they relate to embryos. Essential to his hypothesis is the a priori reasoning that "the earlier these abnormal vessels begin the greater is the extent of their distribution in the lung." Ergo, . . . "by referring to the embryonic bronchial tree at different stages it is possible to date their origin. Thus, the artery in Beneke's case II (1905) supplies the two sequestrated lungs and arises at 3 mm. . . . In cases II and VII of Pryce it supplies the posterior basal sector and arises about the 10-14 mm. stage" (Pryce,[15] p. 463; see also Fig. 24, Pryce and others[16]). This deduction seems quite plausible until one examines the topography of a 13 mm. embryo (the lungs of which are shown in Fig. 4). Then it appears that an aberrant artery, stemming from the aorta just above the diaphragm, must have had to arise at about the level of the seventh thoracic vertebra in order to maintain its relation to the downward migrating celiac artery (see table of positions of celiac artery in human embryos, Evans,[8] p. 648). Second, it would have had to grow up through differentiating tissues to about the level of the fourth thoracic vertebra, in order to invade the hilum of the lung and descend through the lower lobe to reach the posterior basal bronchi. This is unrealistic. But, accepting the hypothesis for the moment, why then should it not have captured the whole lower lobe?

Another untenable embryonic premise is presented in the article of 1947 (Pryce,[16] p. 23). There the authors state that "the reason it is considered that traction by such delicate structures as embryonic blood-vessels, which are essentially capillary, can lead to amputation of bulbous tips of the developing bronchial tree is that the latter are also fragile, being composed only of thin epithelium embedded in mesenchyme." Unfortunately this is far from being a correct description. Even in very early stages the "bulbous tips" are massive bulbs consisting of a thick pseudocolumnar epithelium containing several rows of nuclei (cf. Streeter,[20b] Fig. 4, p. 146), and they never become less massive

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right lung of a 7-year-old white girl. These vessels of aortic origin entered the mediastinal surface of the lung at different levels. The lung had the structure of a left lung and it was drained by an anomalous pulmonary vein which entered the inferior vena cava. When injected, with the superb technique developed by Dr. Liebow, the systemic arteries were found to supply portions of the lingular segment, the medial-, anterior-, and lateral-basal segments, and all of the posterior basal segment. The rest of the lung was supplied by the definitive pulmonary artery. All of the accessory arteries, except the lingular, swept laterally from the posteroinferior angle of the lower lobe. As they approached the bronchi "the branches of the artery became *recurved*, proceeding laterally in close relationship with the bronchi, as is typical of pulmonary arteries. None of these had the spiral anastomosing characteristics of a bronchial artery, despite their aortic origin. There was no overlap in the territory supplied by the two sets of arteries." No cysts were present in this lung.

(Figs. 3 and 6) until the glandular period of growth is completed. How, then, is it mechanically possible for such bulbs to be "captured" by vessels that "are essentially capillary"?

A third source of confusion is the belief that there is competition for the lung buds on the part of the two arteries (Pryce and others,[16] p. 22); or that the persistence of the systemic artery is due to some breakdown in the formation of the pulmonary artery (Smith[18]). What really happens is that very early in development the trachea, the paired bronchial buds, and the esophagus are surrounded by a visceral plexus of capillaries. Soon this system is tapped by incipient pulmonary arteries and veins and by any aberrant systemic channel (arterial or venous) which happens to be present. It is the persistence of these latter connections which produces the varied vascular anomalies reported in the literature. When such an accessory arterial channel persists, it merely takes over the bronchial buds in its vicinity while the incipient pulmonary artery takes over the rest. These zones may then be moved apart by interstitial growth of the larger bronchi so that a "no-man's land" is left between them.* Neither artery has failed or overcome the other; each merely restricts itself to the bronchi with which it is in contact. Meanwhile, the proximal portions of both the typical and the accessory pulmonary arteries begin to develop smooth muscle coats—dense tunicae mediae which distinguish them as arteries. But this zone of differentiation always remains a certain distance behind the growing bronchial buds. Presumably also, in time, the wall of the systemic artery may develop a wall like that of the pulmonary artery which is predominantly composed of elastic tissue.

Finally, there are the numerous reasons why intra- and extrapulmonary sequestration should be considered as having a different etiology. These have been covered so thoroughly and critically by R. Abbey Smith[18] that this ground need not be harrowed again.

How then may the frequent concurrence of lung cysts and systemic pulmonary arteries be explained? A very attractive theory is the one presented by R. Abbey Smith[18]—namely, that after birth that portion of the lung which is subjected to systemic blood pressure may undergo cystic and fibrous degeneration. The strongest evidence that can be summoned in support of this theory is Potter's observation[13] that intralobar sequestration has not yet been detected in 10,000 post-mortem examinations of children under one year of age. But, as Dr. Potter would be the first to point out, failure to demonstrate a condition in innumerable autopsies does not establish the fact that it cannot occur. It merely means that if it does occur it is extremely uncommon. Also, it is probably that conditions which are not immediately lethal seem to be recorded less often in autopsies than one would anticipate from their occurrence in the general population. Finally, Smith's theory would require explanation of the absence of associated cysts in mature individuals in which there is a stout systemic pulmonary artery (e.g., cases by McCotter,[11] Pryce,[15] Kinsella,[10] among

*Later, perhaps, this interarterial zone may be bridged by bronchial arteries and their anastomotic connections with the pulmonary arteries.

others) or in cases where there are five or six aortic pulmonary arteries supplying peripheral branches of the lingular and basal segments (Halasz and associates[9]). Indeed, it is quite possible—witness the 57 cases collected by Cole, Alley and Jones[6]—that systemic arteries to the lower lobes are more numerous than has been suspected, and that many are never seen, surgically, because they are not associated with diseases of the lung or other circumstances requiring surgical intervention.

Because of these considerations, the writer suggests that until there is evidence to the contrary one must fall back upon the theory of coincidental occurrence of lung cysts and systemic pulmonary arteries. An analogy to this may be found in cases of agenesis of the lung. This lesion can be produced experimentally in fetal rats by feeding the mother with a diet deficient in vitamin A. Frequently associated with this condition are anomalies of the aortic arches (Wilson and Warkany[25]). Yet either condition can be present without the other.

SUMMARY

Lung cysts developing spontaneously in a 31 mm. human embryo, and a systemic pulmonary artery supplying the posterior basal segment of a 41 mm. fetus have been discovered in sectioned human embryos of the Carnegie Collection. These anomalies have been described and discussed in relation to Pryce's theory of intralobar sequestration. It is concluded that a hypothesis of coincidental occurrence of these anomalies in certain patients is still all that can be warranted by the facts at our disposal.

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GRANULOMATA OF THE MEDIASTINUM SURGICALLY TREATED AND FOLLOWED UP TO NINE YEARS

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GRANULOMATA of the mediastinum assume surgical significance from two aspects. First, they may present as mediastinal tumors of undetermined etiology for which exploratory thoracotomy is indicated for diagnostic purposes.[1, 2] Second, they occasionally encroach upon vital neighboring structures and cause complications involving the trachea, bronchus, lung, esophagus, or superior vena cava.[3-10]

In the early reports, these lesions were designated "tuberculomata."† [1, 11] Actually, they are conglomerate caseous granulomata of lymph nodes pathologically, often described "consistent with tuberculosis." Although grossly and histologically they are typical of tuberculosis in most of the cases, the etiology cannot be determined with accuracy because organisms are often not demonstrable in the lesions. Fungi are also know to produce similar lesions. A more accurate designation is therefore "mediastinal granuloma."

There have been several reports of these granulomatous mediastinal tumors being removed either entirely or partially. These cases were summarized by Kunkel, Clagett, and McDonald,[12] and they reported 32 cases proved at thoracotomy including 16 of their own. Subsequently, Perdue and Guilfoil[13] reported 3 additional cases. Although these granulomata are considered to be of tuberculous or fungus origin, there is no long-term follow-up of these patients to indicate that they remain free of further manifestations of active disease for significant lengths of time. This report deals with 19 patients who were subjected to exploratory thoracotomy for mediastinal tumors which proved to be granulomata. They have been followed for periods up to 9 years.

Clinical Aspects.—The majority of these patients were discovered to have mediastinal masses on routine roentgenological examination of the chest. Two patients reported having had some discomfort in the chest. One patient had pain in right shoulder and right arm (Case No. 14). Two patients had a slight

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†Tuberculomata are firm, discrete, round lesions. On section they may present a laminated appearance with concentric rings alternating with areas of calcification. Microscopically, one invariably finds areas of caseation. Should they present an area of central cavitation, they should not be designated as true tuberculomata.

TABLE I. PATIENTS WITH MEDIASTINAL GRANULOMATA SUBJECTED TO SURGERY

CASE NO.	SEX AGE	SKIN TESTS	LOCATION AND SIZE	SURGERY	PATHOLOGY	POSTOP. THERAPY	FOLLOW-UP	TIME FOLLOWED (YEARS)
1	E.W. L. M 20	T - 0 H - – C - –	4 cm. rt. ant. sup. mediastinal	Excised	Cystic caseous granuloma of node	Bed rest 5 mo.; no drugs	Asymptomatic	7
2	F. W. B. M 19	T - 0 H - 0 C - 0	7 × 5 cm. rt. post. sup. mediastinal	Excised	Caseous calcified granuloma of node	SM 6 mo.	Asymptomatic	5
3	W. E. H.* M 25	T - + H - – C - –	3 cm. rt. paratracheal	Excised	Tuberculosis of node; *positive* for AFB (culture)	3 mo. bed rest; SM and PAS 42 days	Asymptomatic	6
4	D. P. G.* M 21	T - 0 H - – C - –	3 cm. rt. paratracheal	Excised	Caseous granuloma of node	Bed rest; SM and PAS 3 mo.	Asymptomatic	6
5	M. A. O'B.* M 24	T - + H - – C - –	5 × 3 × 2 cm. rt. paratracheal	Excised	Caseous granuloma; *positive* for AFB (smear)	Bed rest 3 mo.; no drugs	Asymptomatic	9
6	E. L. D. M 23	T - + H - + C - 0	3 × 4 cm. rt. paratracheal	Excised	Fibrocaseous granuloma	None	Asymptomatic	5
7	S. L. R. M 19	T - + H - 0 C - 0	5 × 5 cm. rt. paratracheal	Lesion eroded into lung; incomplete excision of mass and wedge resection of lung	Fibrocaseous granuloma	SM and PAS; bed rest 6 mo.	Asymptomatic	4
8	D. M. K. M 24	T - + H - 0 C - 0	2 5 cm. defect in esophagus at level of carina	Excised from carina and esophagus	Tuberculosis of node; *positive* for AFB (culture)	SM and PAS; bed rest 8 mo.	Asymptomatic	4
9	A. L. F 21	T - + H - 0 C - 0	3 × 4 cm. left hilar	Biopsy	Tuberculosis of node; *positive* for AFB (culture)	SM and PAS; ambulatory, 1 yr.	Asymptomatic; mass regressed in size	2½
10	B. G. M 28	T - 0 H - –	5 × 3 cm. rt. paratracheal	Excision	Fibrocaseous granuloma; calcium	SM-INH-PAS; ambulatory, 1	Asymptomatic	2

10	B. G. M 26	T - 0 H - – C - –	5 × 3 cm. rt. paratracheal	Excision	Fibrocaseous granuloma; calcium	SM-INH-PAS; ambulatory, 1 yr.	Asymptomatic	2
11	M. S. M 48	T - + H - + C - 0	5 × 3 × 8 cm. rt. paratracheal	Excision	Granuloma	INH-SM; ambulatory, 1 yr.	Asymptomatic	1
12	A. C. V. M 21	T - 0 H - + C - 0	3 × 4 cm. rt. paratracheal	Partial excision	Caseous granuloma, lymph nodes	SM-PAS 1 yr.; ambulatory	Asymptomatic	1½
13	J. J. P.* M 26	T - + H - – C - –	4 × 6 cm. left heart border	Mass excised from left pericardium	Cystic granuloma	SM and PAS 42 days; bed rest 6 mo.	Asymptomatic	7
14	C. M.* M 35	T - + H - – C - –	Rt. paratracheal	Partially excised	Fibrocaseous granuloma	SM and PAS 4 mo.	Asymptomatic	6½
15	W. J. S. M 19	T - + H - 0 C - 0	Rt. paratracheal	Excised	Fibrocaseous granuloma	None	Bilat. apical TBC. 3 mo. postop.; then treated with SM-PAS and bed rest for 8 mo.; now asymptomatic	4½
16	F. B. S.** M 35	T - + H - 0 C - 0	Midmediastinal, subcarinal	Unsuccessful removal on rt.; removed from left at 2nd operation	Fibrocaseous granuloma	None	Developed cavitary TBC. RUL 4 yr. postop.; under treatment	4
17	J. C. R. M 29	T - + H - – C - –	2.5 cm. rt. paratracheal	Excision	Fibrocaseous lymphadenitis	None	Asymptomatic	4
18	M. L. M F 22	T - 0 H - 0 C - 0	3.5 × 2.5 cm. rt. treacheal	Biopsy	Granuloma, lymph node	None	Asymptomatic	2
19	H. M. M 27	T - 0 H - + C - 0	4 × 3 cm. rt. paratracheal	Incomplete excision	Granuloma, histoplasmosis	None	Asymptomatic	2½

Key to abbreviations: T = Tuberculin; H = Histoplasmin; C = Coccidioidin; + = Positive; 0 = Negative; – = Not reported.
*Reported by Storey and Lyons.[3]
**Reported by Lyons and Storey.[4]

daily elevation of temperature for 3 weeks prior to admission. Four patients complained of cough, one with hemoptysis, several years previously. One patient (Case No. 15) complained of fever, malaise, and weight loss (subsequently developed overt pulmonary tuberculosis 2 months postoperatively). One patient (Case No. 17) was placed on streptomycin, isonicotinic acid hydrazide (INH), and para-aminosalicylic acid (PAS) therapy after biopsy of a mediastinal tumor showed granuloma; the mass increased in size roentgenologically over the next several months and excision was performed. One patient, who had a partial esophageal obstruction due to granuloma, had dysphagia for 2 years.

Physical examination was noncontributory in each case. Hematological examination, urinalysis, and the other usual laboratory examinations were not helpful in establishing the diagnosis.

Skin tests for tuberculosis, coccidioidomycosis, and histoplasmosis were performed in many of the cases. The results of these tests are tabulated in Table I.

Bronchoscopy was performed in many of the cases. No endotracheal or endobronchial lesion was demonstrated, but extrinsic compression of the trachea was present in one case. Esophagoscopy was negative in the case in which esophagram revealed an extrinsic mass encroaching upon the esophagus.

In none of these cases was there any concomitant lesion visible on the roentgenogram or evident on physical examination which made it possible to conclude that the mediastinal mass was definitely tuberculous or fungous in origin and not a mediastinal tumor of other type.

Roentgenological Aspects.—The majority (15) of these mediastinal granulomata presented as mediastinal masses projecting from the mediastinum to the right above the hilum of the right lung (Fig. 1). Typically, they were situated in the right paratracheal region, were usually lobulated, and occasionally revealed spotty calcification. Tomography was useful in delineating the size, position, and character of these lesions. One of the masses was situated beneath the carina of the trachea, one at the left hilum, and one at the left heart border. One case presented a mediastinal tumor encroaching upon the esophagus, suggesting the possibility of a leiomyoma of the esophagus (Fig. 2). These masses ranged in size from 2.5 cm. to 8 cm. in the largest dimension. In none of these case was there significant hilar adenopathy (one presented as a left hilar mass). In no case was a lesion noted in the lung fields. In one patient (Case No. 14) obstruction of the right subclavian vein was demonstrated by angiography.

Anatomic Aspects.—Fifteen of these granulomata presented in the right paratracheal region beneath the mediastinal pleura, immediately superior to the right hilum. They usually were situated superior to the azygos vein and anterolateral to the trachea, somewhat posterolateral to the superior vena cava. Very often the mass was intimately adherent to the trachea, bronchus, superior vena cava, azygos vein, or pulmonary artery. Occasionally additional

enlarged nodes were felt at or below the hilum of the lung. The lung was frequently adherent to the mass, but it was involved in the mass in only one instance.

The granuloma presented at the left heart border, the subcarinal region, encroaching upon the esophagus from the right just below the bifurcation of the trachea, and at the left hilar region, in one instance each.

Pathologic Aspects.—Grossly, these lesions were lobulated and either solid or cystic. They were hard in some areas and soft in others. Many of the cysts ruptured easily (Fig. 3, *B*). Gross calcification was present in 3. The central portions of these granulomata were yellow or white and contained caseous material of a chalky or cottage-cheese consistency (Fig. 4).

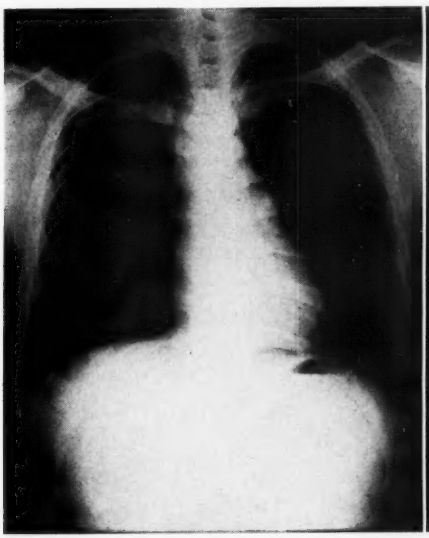


Fig. 1.

Fig. 2.

Fig. 1.—Posteroanterior chest roentgenogram of patient with mediastinal granuloma. Typical roentgen finding of lobulated mass projecting to right of superior mediastinum.

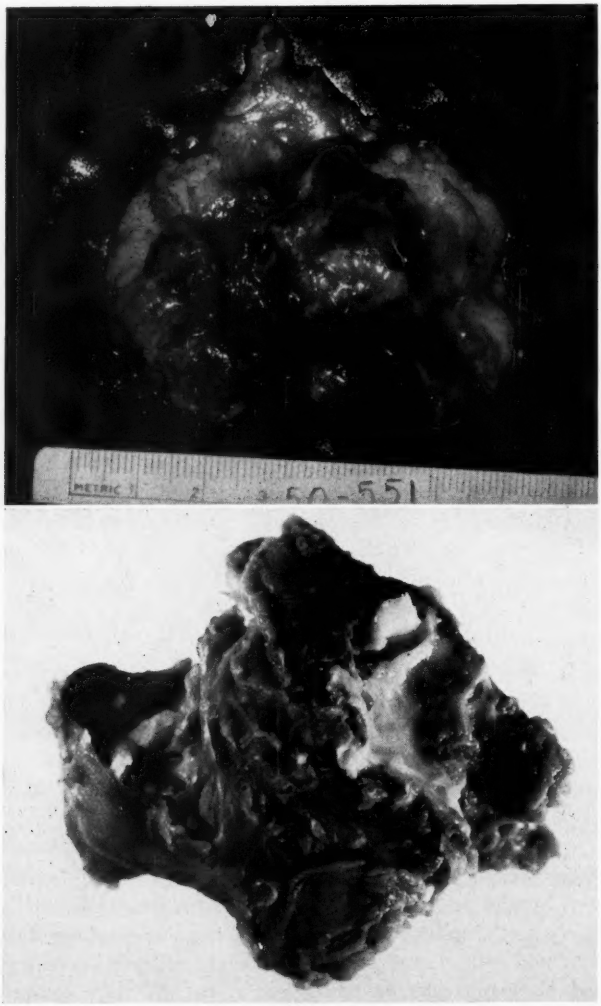
Fig. 2.—Esophagram showing granuloma of mediastinum causing filling defect in esophagus.

All of these lesions were found to be granulomata histologically. Some were described by the pathologist as "fibrocaseous granulomata" and others described simply as "granulomata." Some of the lesions showed small tubercles with epithelioid cells, Langhans' type of giant cells and caseation (Fig. 5). Some revealed no giant cells or epithelioid cells, but were composed mainly of fibrous tissue and inflammatory cells. Nine of these granulomata had remnants of lymph nodes in the walls. The remaining 9 had no lymph node component recognizable grossly or histologically.

All of the lesions were examined microscopically for acid-fast bacilli and 18 were cultured for *Mycobacterium tuberculosis*. Three of the lesions were positive for *Mycobacterium tuberculosis* on culture. One which was negative

on culture had been positive for acid-fast bacilli microscopically. Many of the granulomata were examined microscopically and cultured for fungi, and one was positive for *Histoplasma capsulatum.*

A.



B.

Fig. 3.—*A*, Gross appearance of typical mediastinal granuloma.
B, Gross appearance of a mediastinal granuloma which was partially opened during excision.

Surgical Aspects.—Sixteen of these lesions were approached through a right posterolateral thoracotomy and 2 through a left posterolateral thoracotomy. One was approached at a first stage by right thoracotomy, and because

Fig. 4.—Cut gross specimen of mediastinal granuloma in Fig. 3, *B* showing cystic nature and caseous central portions.

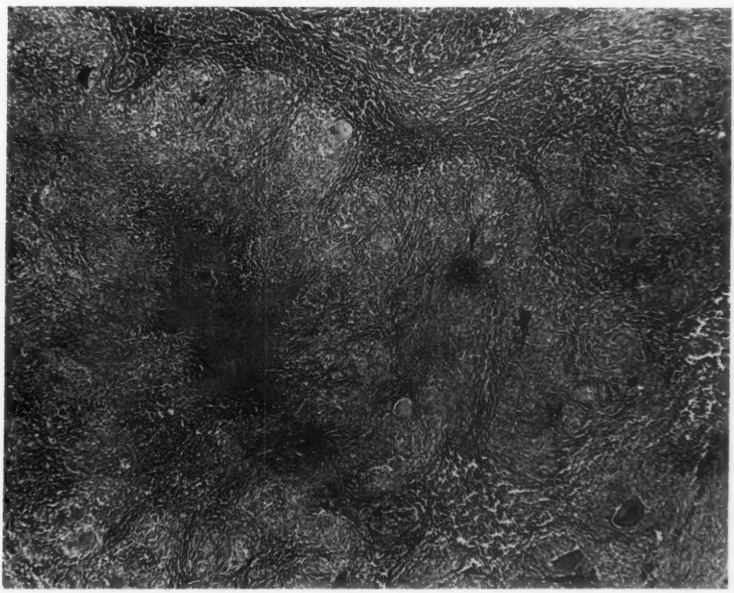


Fig. 5.—Photomicrograph of typical mediastinal granuloma (×50). Giant cells, epithelioid cells, and caseous necrosis are present.

it was impossible to remove the subcarinal mass at the first operation, a left thoracotomy was performed later to complete the removal. Posterolateral thoracotomy through the fourth interspace gave excellent exposure of the typically placed paratracheal mass. The azygos vein was divided in most cases in which excision of the mass was performed.

Upon exposure of the mass, it was found to be injudicious to attempt removal of the granuloma completely in 6 cases because of marked adherence to the trachea, bronchus, vena cava, or pulmonary artery. Therefore, biopsy only was performed in 2 cases and partial excision of the mass was performed in 4 cases. In the remaining 13 cases, the mass was removed completely. In a number of these cases, the mass was opened during the course of removal and the caseous material was spilled into the thoracic cavity. In one case, the right paratracheal mass was intimately adherent to the right upper lobe of the lung and a wedge of right upper lobe was removed along with a portion of the lesion. Partial excision of the mass in Case No. 14, in which obstruction of the right subclavian vein was shown preoperatively, relieved the venous obstruction.

Postoperative Complications and Treatment.—All of these patients recovered from their thoracotomy without complication. There was no case of empyema or pulmonary infiltration recognizable postoperatively (except for Case No. 15). Eleven patients received some combination of streptomycin, INH and/or PAS for periods ranging from 6 weeks to one year with no postoperative evidence of disease during follow-up periods up to 6 years. Two patients received bed rest only in periods ranging from 3 months to 5 months, and these patients are all well in follow-up periods up to 9 years. Six patients received no postoperative therapy directed toward the prevention of complications or exacerbations of tuberculosis, and 2 of these patients have subsequently developed active pulmonary tuberculosis. One of the patients that received no particular postoperative therapy was demontrated to have *Histoplasma capsulatum* organisms in the specimen, and he has been well for 2½ years following biopsy of the mass.

Follow-up of These Cases.—These cases have been followed up to 9 years, and all have been asymptomatic without evidence of further disease except 2 patients.

Case No. 15 had a history of malaise, fever, night sweats, and a 20-pound weight loss for one month prior to admission. Physical examination revealed no abnormalities except for fever of 103° F. Roentgenogram of the chest was negative except for a right upper mediastinal mass. Skin tests for tuberculosis were positive. During hospitalization, the temperature returned to normal and the patient gained 10 pounds. Approximately 6 weeks after admission, thoracotomy was performed with a preoperative impression of Hodgkin's disease, and the mediastinal mass was excised. Small nodules were felt deep in the substance of the lung. No biopsy of these was taken because of the depth of the lesions in the lung. The specimen was a fibrocaseous granuloma, and smears and cultures for tuberculosis were negative. Antituberculosis therapy

was not given. The postoperative course was uneventful, and the patient was discharged 4 weeks postoperatively. While at home he began to have fever and malaise again 2 months postoperatively and was readmitted to the hospital with bilateral apical pulmonary tuberculosis. Treatment with streptomycin and PAS was then instituted and continued for 8 months. He is asymptomatic at present.

Despite the fact that there was no x-ray or bacteriologic evidence of pulmonary tuberculosis preoperatively, it may be that this patient was suffering from active tuberculosis at the time of thoracotomy. In any event, his resistance to tuberculosis was insufficient to prevent the development of bilateral apical pulmonary lesions. Postoperative antituberculosis chemotherapy may have prevented the development of clinically active pulmonary tuberculosis in this case.

Case No. 16 had a large subcarinal mass removed through a left thoracotomy after previous right thoracotomy had not given adequate exposure for removal of the mass. He received no bed rest or chemotherapy for tuberculosis postoperatively. The lesion pathologically was nonspecific fibrocaseous granuloma and no acid-fast bacilli were demonstrated by smear or culture. The postoperative course was uneventful and the patient felt entirely well for 4 years after operation. He developed cavitary pulmonary tuberculosis in the right upper lobe with positive sputum 4 years postoperatively.

This patient most likely has less than average constitutional resistance to tuberculosis and postoperative chemotherapy would probably have suppressed latent foci of tuberculosis sufficiently to prevent the subsequent development of cavitary pulmonary tuberculosis.

DISCUSSION

Our experience with the excision of these granulomata is similar to that reported by others. Pathologically, most were chronic fibrocaseous granulomata. Four were positively identified as tuberculosis and one as histoplasmosis by demonstration of organisms. These masses are generally assumed to develop as cystic caseous enlargement of lymph nodes, and 9 of these granulomata exhibited remnants of lymph node histologically. The remainder probably represent complete destruction of the nodes or may be chronic mediastinal abscesses which develop following rupture of a caseous lymph node, such as reported by Samson.[14]

Exploratory thoracotomy is indicated for all mediastinal tumors of undetermined etiology. Complete removal of a mediastinal granuloma is the procedure of choice if this can be accomplished. Removal of the granuloma is indicated to prevent complications of encroachment of the granuloma on the trachea, bronchus, superior vena cava, esophagus, or lung and to remove a focus of possible exacerbation of disease. If complete excision is impossible because of adherence to neighboring vital structures, removal of as much of the granuloma and the caseous contents as is feasible will reduce the size of the mass and probably is helpful in preventing further complications involving

these structures. However, because most of these granulomas do not cause serious complications, biopsy alone is sufficient if further excision appears to be contraindicated.

Mediastinal granulomata of tuberculous origin are probably manifestations of postprimary localized mediastinal lymphadenitis. The development of clinical pulmonary tuberculosis in 2 of these 19 patients is probably not a result of the surgical intervention per se, but is a manifestation of a constitutional susceptibility of these individuals to tuberculosis. These individuals may develop exacerbations at any time, since they harbor latent tuberculous foci. Medlar[15] has emphasized that caseous foci contain viable tubercle bacilli that may cause a relapse of disease years later. Because the 2 patients who developed clinical pulmonary tuberculosis did not have antituberculous chemotherapy, we now assume that all of these granulomata which are not demonstrated to be of fungus etiology are of tuberculous etiology although organisms are not demonstrated. Therefore, all of these patients in whom a fungus is not demonstrated in the lesion receive antituberculous chemotherapy routinely for 6 to 12 months. The chemotherapy is given to prevent possible sequelae of contamination of the pleural cavity with the contents of the granulomata during removal and to prevent further exacerbations elsewhere. We prefer to give INH and PAS and reserve streptomycin for possible further use in the future.

CONCLUSIONS

1. Exploratory thoracotomy is necessary for the diagnosis of mediastinal granulomata when there is no other clinical evidence which would indicate the granulomatous nature of these lesions.

2. Removal of a portion of the granuloma for biopsy is essential in every case. Excision of as much of the mass as is feasible without undue risk is accompanied by no further complications than biopsy alone.

3. Postoperative antituberculous therapy for 6 to 12 months should be administered to prevent subsequent spread or reactivation of tuberculosis unless a nontuberculous etiology is definitely established.

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TUMOR-FORMING AMYLOIDOSIS OF THE LUNG

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INCOMPLETE knowledge of the etiology and nature of amyloid degeneration makes classification of amyloid disease difficult. In 1929, Lubarsch[8] suggested a uniform nomenclature and coined the term atypical amyloidosis for those cases deviating from the well-known classical type. Herxheimer and Reinhart[4] suggested a division of amyloidosis into two types, namely generalized and local, while Rosenblum and Kirschbaum[11] classified the condition as primary or idiopathic and as secondary or symptomatic with subdivision of each group into diffuse (typical) and local (atypical) forms. King[5] based his classification on the anatomic distribution and distinguished typical and atypical amyloidosis associated or not associated with other disease. The classification often referred to in the literature is that suggested by Reimann and associates.[10] They recognized three groups in addition to the secondary amyloidosis of chronic infectious diseases, namely, primary atypical amyloidosis, characterized among other things by the absence of any disposing basal disease; nodular, solitary or multiple local amyloidosis (amyloid tumors) and, finally, amyloidosis associated with multiple myeloma. Though nodular amyloidosis is usually primarily atypical, it appears justified to assign amyloid tumors to a special group because of their characteristic pathologic features.

Amyloid tumors have been the subject of research for many years, especially in Germany. Leupold[6] emphasized the value of publishing cases of localized amyloidosis and stressed the importance of descriptions of the tumors in the elucidation of problems bearing on amyloidosis. The factors considered of importance for the development of secondary amyloidosis can nearly always be excluded in nodular amyloidosis. Another essential difference is the fact that amyloid tumors are seldom seen in parenchymatous organs. They show a predilection for striated or smooth muscle, skin, lymph nodes, and, occasionally, for the mucosa of the digestive and urinary tracts. The base of the tongue is also a common site of circumscript nodular amyloidosis. Multiple amyloid tumors in the upper respiratory tract are not uncommon. This form of amyloidosis is rarely seen in the lung parenchyma, and a search of the literature failed to reveal reports of more than 13 cases. All of them have been recently reviewed (Glauser[2]). The pulmonary changes sometimes produced symptoms of pneumonia and pleurisy, sometimes they produced no clinical symptoms and were discovered incidentally at roentgen examination or post

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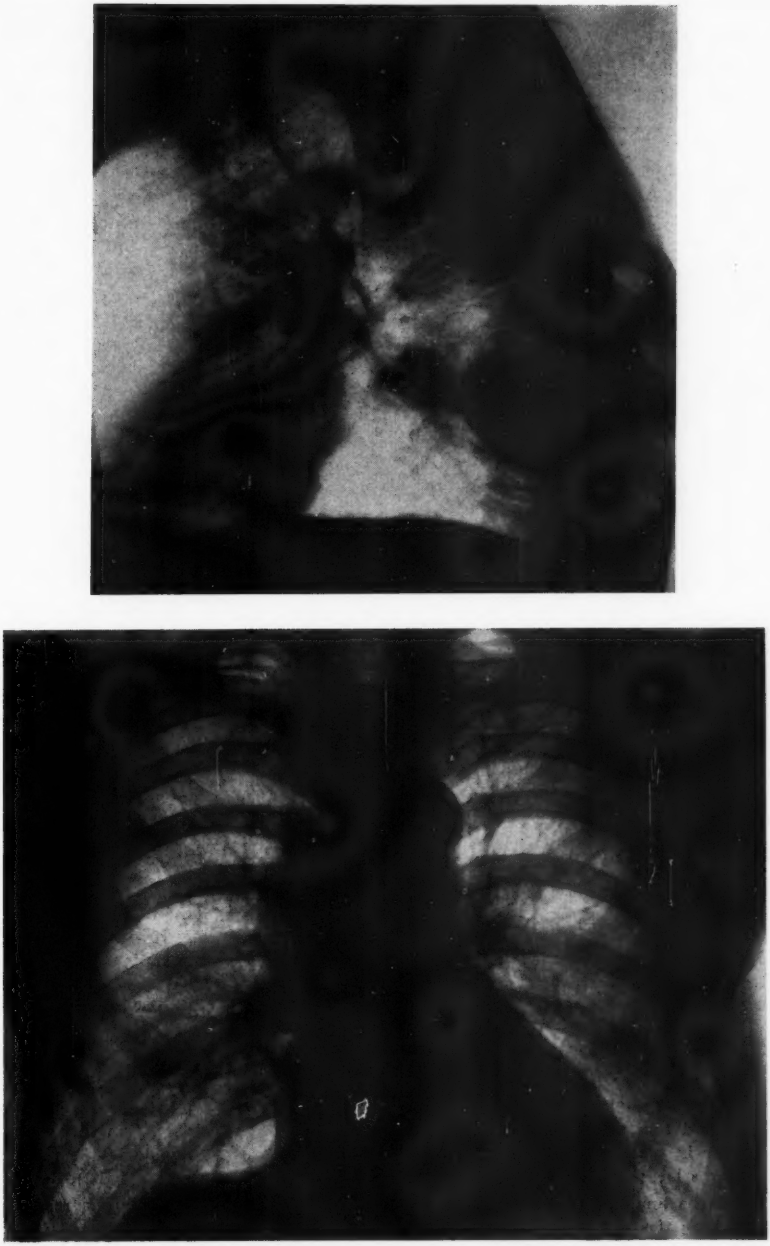

Fig. 1.—Frontal and lateral views showing 3 of the 4 tumorlike processes in the left lung.

mortem. In one case of solitary amyloid tumor, the patient, a man aged 66 (Haynes and co-workers[3]), was operated on because of a presumptive diagnosis of pulmonary tumor. In the case reported below the preoperative diagnosis was secondary lung tumor from a primary tumor elsewhere.

CASE REPORT

K. A., a farmer aged 67 years, was treated conservatively for duodenal ulcer in 1932 and 1933. In 1955, the patient sustained a blow against the left flank with costal infraction (site no longer demonstrable in roentgenogram), otherwise he had always felt well.

He was admitted to the department of thoracic surgery on Aug. 29, 1956, after repeated mild infections of the respiratory tract during the previous 3 to 4 months and a markedly progressive nonproductive cough for 5 weeks. An earlier roentgenogram of the chest (August 17) had shown the presence of at least 3 tumorlike changes in the left lung (Fig. 1). Of these, 2 were located in the lower lobe and one in the upper lobe.

Apart from the troublesome cough, the patient felt well. Routine examination revealed no pathologic palpatory changes of the internal organs or lymph nodes and roentgen examination of the digestive tract, urinary tract, and skeleton showed nothing abnormal. The blood values and the blood picture were also normal. The sedimentation rate was 16 mm. per hour.

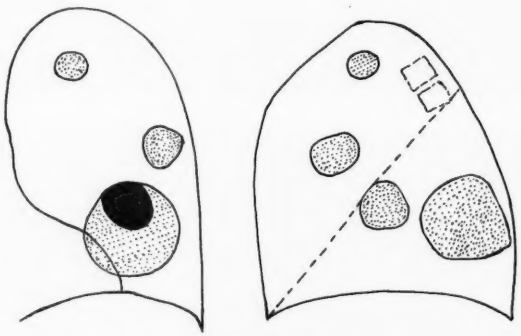

Fig. 2.—Schematic drawing of the localization of the processes as seen at exploratory thoracotomy.

Bronchoscopy showed nothing abnormal, and histologic examination of the bronchial secretion contained no cells ascribable to a malignant process. However, since there was still reason to suspect that the roentgenographic lung changes were malignant, exploratory left-sided thoracotomy was performed on Sept. 14, 1956.

No pleural changes were observed. However, a flat, well-defined and slightly nodular tumor, the size of a billiard ball, was palpated at the border between the apical and basal segments of the lower lobe and just below the visceral pleura. Adjacent to this lump was a similar tumor the size of a walnut, which extended to the oblique fissure. In adjacent parts of this apical segment of the lower lobe were smaller indurated foci. Two more walnut-sized tumors of the same nature were felt in the upper lobe. One was located in the upper lingual segment and the other, which was not demonstrable in the roentgenograms, was located in the anterior subsegment of the apical segment (Fig. 2).

All the tumors felt alike: they were firm to hard and surrounded by macroscopically normal lung tissue. The smaller of the two lower lobe tumors was enucleated for rapid histologic diagnosis. The microscopic picture was not suggestive of malignant tumor. The process was of homogeneous character and contained abundant amyloid deposits. On

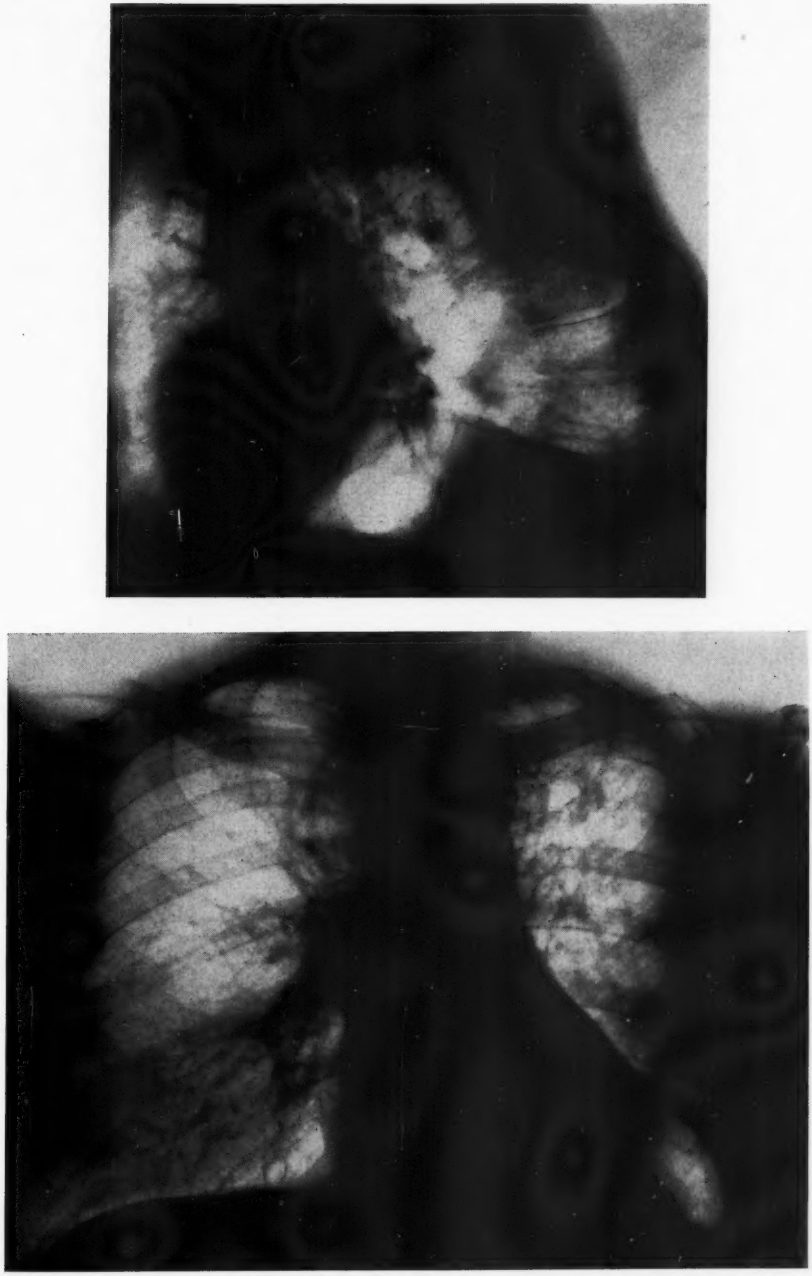

Fig. 3.—Roentgenograms 4 months after operation (Jan. 8, 1957).

receipt of the pathologist's report, the operation was continued with removal of the lower lobe and enucleation of the two upper lobe lumps. Nowhere had the tumor involved the large bronchial branches.

The postoperative course was uneventful, and 14 days later the patient left the hospital (Sept. 29, 1956). During the first few weeks convalescence was smooth, and the patient had no respiratory tract symptoms. However, he fell acutely ill on Oct. 12, 1956, with a virus infection of influenza type, and 2 days later expectoration was abundant and sometimes sanguinolent. The patient was readmitted to the department of thoracic surgery on October 16. Roentgen examination of the lungs showed an intrapleural space partly filled with fluid and located at the earlier site of the enucleated tumor in the apical segment of the upper lobe. Intense antibiotic therapy controlled the fever, and the initially abundant expectoration ceased. Three weeks later, slight albuminuria and microscopic hematuria were noted. These signs were still persistent when the patient left the hospital in an otherwise good condition on Dec. 1, 1956. Since then the patient has had no respiratory tract symptoms (Fig. 3).

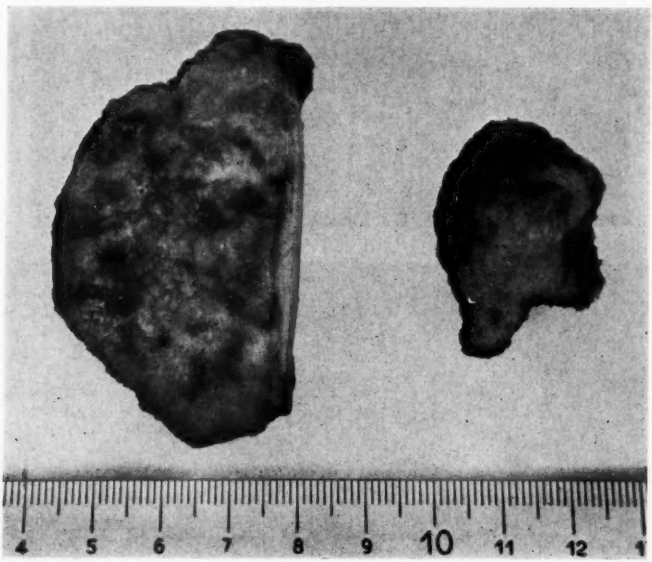

Fig. 4.—Gross specimens from the large tumor in the lower lobe and from one of the tumors in the upper lobe. The cut surfaces were waxlike with scattered ill-defined darker portions.

Supplementary Examinations.—Because of the histologic picture of the processes, the blood and urinary proteins were repeatedly determined, but without providing any support for a diagnosis of myeloma. Further roentgen examinations of the skeleton were also negative. Electrophoretic analysis of proteinbound blood carbohydrate showed normal values. The blood-forming bone marrow was normal. Further examination, because of persistent renal symptoms, suggested the possibility of glomerulonephritis. The patient is still under observation.

Macroscopic Examination.—The specimen consisted of the entire left lower lobe and 2 tumors from the upper lobe. The largest diameter of the tumor in the lower lobe was 6 cm. The other tumors were barely the size of walnuts. The tumors were well defined against the surrounding lung parenchyma, which was markedly compressed at several sites and the tumors were enucleated without difficulty. The tumors were firm or hard and the

surface was finely nodular. The cut surface was gray-white to gray-yellow and vitreous, but in some places granular with scattered, chalky spots. In addition, diffusely outlined gray-black foci, anything up to the size of confetti, were noted (Fig. 4). The surrounding lung parenchyma was aerated, rich in blood, and the site of moderate anthracosis. The

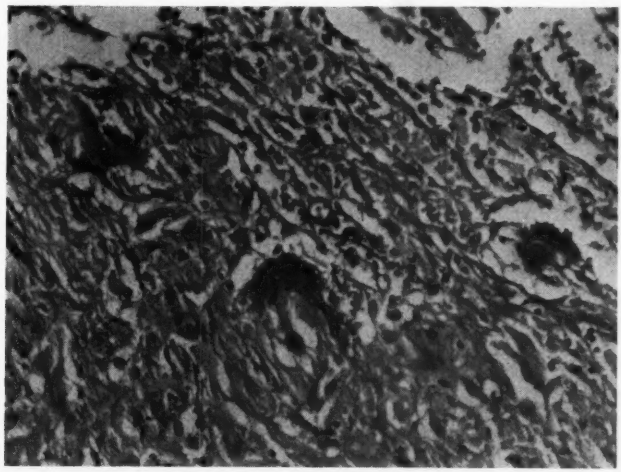

Fig. 5.—Multinuclear giant cells in the periphery of amyloid deposits.

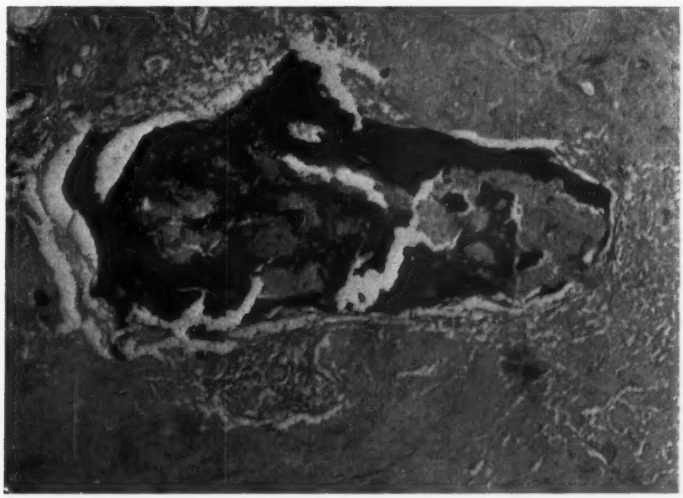

Fig. 6.—Dystopic bone formation in amyloid masses.

large bronchi adjacent to the large tumor in the lower lobe were slightly dislocated by the lesion, but macroscopically they did not appear to be in communication with the actual tumor. The bronchi were slightly widened and the lumina contained a small amount of mucus. In the periphery, the bronchi showed mural thickening, but the mucosa was intact everywhere. Some pea-sized lymph nodes were found in the hilum; they were firm, and the cut surface was anthracotic.

Histologic Examination.—The tumorlike deposits in the lung were built up of closely packed homogeneous trabeculae and laminae, poor in cells, between which were thin streaks of connective tissue with spindle-shaped cells. In large regions the tissue was loose and granular. Between the trabeculae were small deposits of carbon. Adjacent to

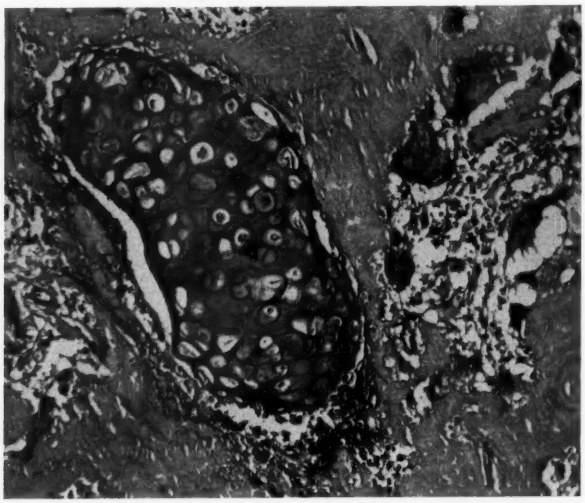

Fig. 7.—Deposit of multinuclear giant cells and round cells in slit-shaped spaces adjacent to a cartilaginous island in the middle of the large lower lobe tumor.

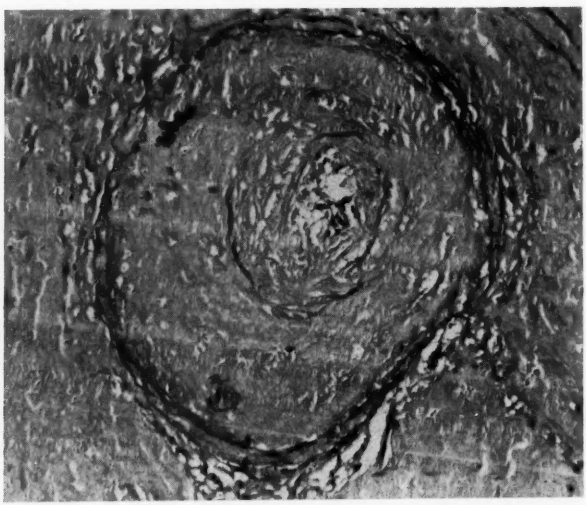

Fig. 8.—Artery with amyloid deposits in all layers of the wall and pronounced narrowing of the lumen.

and often radiating from the localized circumscript deposits were other deposits in alveolar septa and neighboring vessels and bronchi. The alveolar septa were thickened and the capillaries were markedly narrowed. The changes decreased toward the periphery.

In some of the large arteries, all of the layers of the wall contained deposits, and one large bronchus contained foci of hyaline substance in the mucosa, whose epithelium showed squamous epithelial metaplasia.

The deposits stained pink with hematoxylin and eosin. The intensity of the color varied markedly from one portion to another. In preparations stained with van Gieson stain, the deposits were orange and thin, red bands of connective tissue were seen between the coarse trabeculae. Of our most common routine amyloid staining methods, methyl violet often stained the trabeculae bright red, but the degree of metachromasia varied widely in different regions. Congo red stained the hyaline substance red in large regions, but often not strong enough to distinguish from connective tissue and the surrounding lung parenchyma. When stained with Lugol and iodine-sulphuric acid, small regions were seen with an olive-green hue, while the bulk of the masses did not show the color reaction typical of amyloid.

The tumors often showed foreign body giant cells with numerous irregularly arranged, rounded, or oval nuclei and homogeneous cytoplasm, which often contained substance which stained metachromatically with methyl violet. The giant cells often lay in the periphery of the deposits (Fig. 5), but were sometimes seen centrally in the tumors in and close to small slit-shaped spaces around irregularly shaped chalky lamellae and bone trabeculae (Fig. 6). In addition, these regions contained abundant round cells, most of which were plasma cells. In the large tumor in the lower lobe, close to small epithelial-lined spaces (mucus glands), were single regular cartilaginous islands (Fig. 7) probably representing rests of a completely changed bronchus.

In addition to the staining methods for amyloid, the histologic preparations were studied by elastic staining according to Weigert and silver impregnation according to Hortega as modified by King.[5] Frozen sections of the preparations were also stained with scharlach I. In sections stained for elastic tissue, the amyloid assumed a weak, brown-red color. The amyloid masses often contained poorly outlined vessel structures with stratified, round, or oval formations with central endothelial-lined spaces filled with red blood cells (Fig. 8). The lumen was markedly narrowed and the thickened vessel wall contained rests of concentrically arranged elastic lamellae. On silver impregnation, amyloid masses in large regions showed intense argyrophilia, while in other regions they did not take on any stain at all. The amyloid trabeculae often showed small, clear vacuoles which, when stained with scharlach I, took on a clear red color.

DISCUSSION

The case just described agrees with most of those on record and the changes observed fill the criteria usually accepted for a diagnosis of nodular amyloidosis: (1) the macroscopic appearance of the deposited substance and the presence of circumscript foci; (2) typical color reaction with amyloid staining methods but also poor agreement with different methods (Lubarsch[8]), which some authors (Glauser and others[2]) regard as suggestive of chemical differences in the amyloid deposit; and (3) lack of predisposing so-called basal disease.

Whether the amyloid had been deposited also in organs other than the lung in the case under discussion can, of course, not be decided. No clinical symptoms were available to support such an assumption.

Further features regarded as characteristic of nodular amyloidosis are deposits of multinuclear giant cells and the occurrence of bone. Thus, as pointed out by Tsunoda and others,[13] giant cells occur almost regularly in localized amyloidosis but not in generalized amyloidosis. Tsunoda,[13] von Werdt,[14] and others claimed that the cells consist of phagocytic cells of the connective

tissue or the vessel walls, while other authors (W. Fischer[1]) do not believe that giant cells have any phagocytic power but interpret them as a manifestation of a tissue reaction to a foreign substance. However, the fact that, as in the case under discussion, the cells often contain a substance staining like amyloid argues for the cells possessing phagocytic power. In experimental investigation on animals, Ljubimow[7] has shown that amyloid transplanted under the skin is absorbed by giant cells. Then, however, the amyloid absorbed by giant cells often loses its typical color reaction, a loss which has been accepted as a sign of chemical change in the absorbed amyloid.

Calcification and ossification in amyloid tumors have been observed in many cases on record. Small regular cartilaginous islands have also been reported. While the cartilage is probably derived from the bronchi, whose normal structure is obliterated by amyloid deposits, the spongy trabeculae are probably ascribable to an angious bone formation (Scheidegger[12]). The calcification is often accompanied by fatty degeneration of the tumors.

The occasionally abundant deposits of plasma cells in the amyloid masses merit comment. Diffuse or circumscript amyloid deposits are relatively common in multiple myeloma, and then usually with the typical localization of primary amyloidosis and in myeloma foci as well as in and around joints. In the present case, thorough, repeated clinical examinations failed to reveal any definite support for multiple myeloma, and the assumption that amyloid tumors in the present case are due to multiple myeloma and represent extramedullary myeloma foci appears less likely.

SUMMARY

A report is given of a case of multiple nodular amyloidosis of the lung in a man, aged 67. Roentgen examination revealed 3 of 4 rounded formations in the left lung. The lower lobe was removed and the tumors in the upper lobe were enucleated. Histologically, the tumors were built up of homogeneous masses, poor in cells and staining like amyloid. Abundant round cells, mainly plasma cells, were observed as well as multinuclear giant cells and, in some regions, deposits of bone and cartilage.

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LIPOMAS OF THE BRONCHUS

A Presentation of Two Cases and an Analysis of the Literature

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BRONCHIAL lipomas, as well as other benign bronchial neoplasms, formerly pathologic curiosities, have become surgically remedial conditions as techniques for accurate diagnosis and successful removal have been developed.

Kernan,[1] in 1927, was the first to treat a bronchial lipoma by surgical means when he successfully removed one by bronchoscopy. Prior to this time three such lesions had been reported from autopsy material.[2-4] Since Kernan's report, however, 19 additional lipomas or lipofibromas have been treated by bronchoscopy,[5-15] bronchotomy,[16] or where irreparable lung damage was present, by lobectomy[17-19] or pneumonectomy,[20] while only 2 cases have been added from autopsy sources.[21, 22] Thus, the modern management of another disease process capable of destroying the host has developed from knowledge gained through post-mortem studies.

All benign endobronchial tumors fundamentally exert similar influences on the lungs; by partial to complete obstruction of the bronchial lumen, the egress of normal secretions from the bronchial mucosa is obstructed, and inflammation in and around the smaller bronchi and bronchioles occurs with pneumonitis, abscess formation, and/or bronchiectasis as the result.

CASE REPORTS

Case 1.—A 56-year-old white man was admitted to Baylor University Hospital because of a nonproductive, persistent cough of 10 months' duration. Ten months previously he had developed a "respiratory, flu-like infection." Two months before admission he became feverish and an x-ray examination of the chest was reported as showing fibrosis and pleuritis.

Posteroanterior, lateral, and laminographic examinations of the chest (Fig. 1) revealed pneumonitis and atelectasis of the basilar segments of the right lower lobe. A nodularity in the region of the hilus was suggestive of carcinoma.

Bronchoscopic examination demonstrated a rounded, smooth, pulsating, epithelized mass just beyond the superior segmental orifice of the right lower lobe which almost completely occluded the lumen of the basilar segmental bronchus. Biopsy of the pulsating tumor was considered inadvisable.

Thoracotomy by one of us (W. M. A.) revealed the basilar segments of the lower lobe to be markedly involved in suppurative pneumonitis with thick vascular adhesions extending

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Fig. 1.

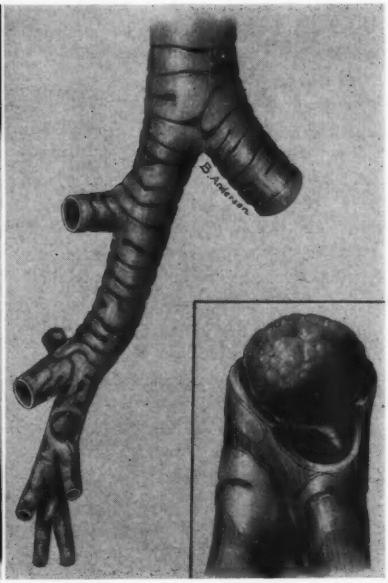

Fig. 2.

Fig. 1.—Case 1. Body section radiograph at 12 cm. which shows mass obstructing RLL bronchus and obstructive pneumonitis.

Fig. 2.—Case 1. Diagram which illustrates relationship of lipoma to RLL bronchus. Wrinkled surface of tumor is shown in inset.

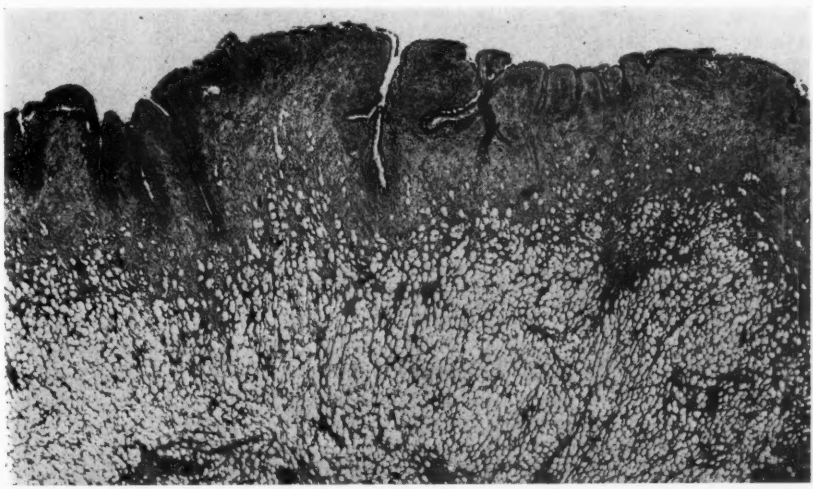

Fig. 3.—Case 1. Lipoma covered by wrinkled surface. Subepithelial tissues scarred and infiltrated by chronic inflammatory cells. (×25; reduced ¼.)

between the visceral and parietal pleura. The hilar and right middle and lower peribronchial lymph nodes were moderately enlarged, soft, and succulent. The upper and middle lobes appeared unremarkable. Accordingly, the right lower lobe was resected.

Pathologic examination: The specimen consisted of a right lower lobe which weighed 335 Gm. A pyriform tumor arose by a narrow pedicle from the wall of the basilar segment and projected proximally to occlude almost the main bronchus of the basilar segment (Fig. 2). The tumor measured 13 by 10 by 10 mm. while its stalk measured 3 mm. in length and 2 mm. in diameter. Its external surface was wrinkled and its interior was of a yellow-gray, solid, rubbery consistency.

The basilar segments of the lobe were atelectatic and nodular while the superior segment was of normal consistency.

Microscopic examination (Fig. 3) disclosed a relatively smooth, intact, respiratory epithelium covering the tumor with frequent epithelial invaginations. In some places the epithelium was only one cell in thickness, while in other places, squamous metaplasia had occurred. The interior was composed of adult fat cells supported by a scanty fibrovascular stroma. A zone of fibrous scar tissue separated the surface epithelium from the interior of the tumor. The pathologic diagnosis was fibrolipoma of the right lower lobe bronchus with chronic obstructive pneumonitis. The patient has been free of symptoms during the ensuing 16 months.

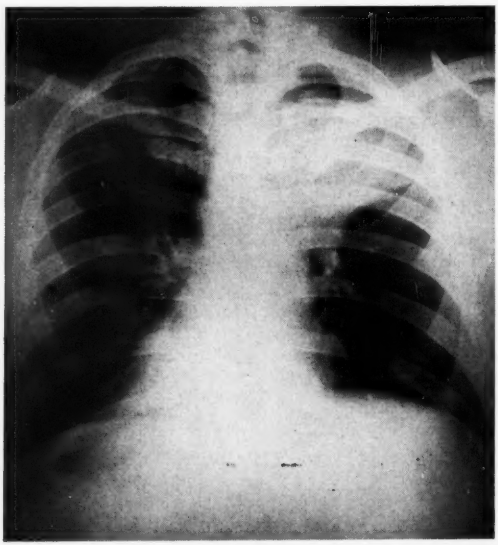

Fig. 4.—Case 2. Atelectasis of RUL which resulted from bronchial obstruction due to lipoma. (AFIP Acc. No. 138325-3.)

CASE 2.—A 32-year-old white man was admitted to the Brooke General Hospital, Fort Sam Houston, Texas, because of a cough of 21 months' duration, accompanied by a loss of 35 pounds of body weight. Pains in the back and right chest had been present for one year. X-ray studies revealed atelectasis of the right upper lobe (Fig. 4). As a biopsy obtained by bronchoscopy was suggestive of anaplastic carcinoma, a right pneumonectomy was performed by one of us (D. L. P.).

Pathologic examination: The pleural surfaces of all lobes were covered by thin fibrinous adhesions. At the origin of the right upper lobe bronchus a polypoid mass filled the lumen and projected into the right main bronchus (Fig. 5). It was attached by a base

measuring 2 by 1 cm. and projected proximally for a distanre of 3 cm. The consistency of the lesion was similar to yellow greasy fat. The upper lobe parenchyma was dense and fibrotic and the bronchi were dilated with mucopurulent material and had thickened walls. The middle and lower lobes were congested and somewhat more fibrous than normal.

On *microscopic examination* the tumor consisted of adult fat tissue covered by respiratory epithelium. All lobes showed chronic pneumonitis with fibrosis, particularly the upper lobe. There was a notable population of eosinophils throughout all lobes. The pathologic diagnosis was polypoid lipoma of the right upper lobe bronchus with bronchiectasis of the right upper lobe and fibrosis and chronic pneumonitis of the right upper, middle, and lower lobes. The patient has had no further difficulties in the 12 years since his operation.

Fig. 5.—Case 2. Polypoid lipoma projecting into right main bronchus. (AFIP Acc. No. 138325-3.)

COMMENT

The stimulus that induces fat which is normally present in the bronchus[19] to undergo neoplastic transformation is unrecognized. Bronchial lipomas most frequently affect individuals in the fifth decade, the average age being 51.1 years among the 20 cases that have been treated surgically, the youngest 29[13] and the oldest 64 years of age.[6] Eighty-eight per cent occurred in men. The duration of symptoms varied from 7 weeks[16] to 15 years[12] with an average duration of 3 years in 13 cases. The symptoms, signs, and radiologic findings were those of bronchial stenosis with accompanying pulmonary parenchymal disease. In several cases the radiographs were suggestive of bronchial carcinoma. The majority of the tumors could be visualized by bronchoscopy. The tumors arose from the bronchial tree in the following locations: trachea, 1; left main, 8; left upper lobe, 3; left lower lobe, 2; right main, 3; right upper lobe, 2; right lower lobe, 2. The composite dimensions of the tumors in the 12 cases treated surgically, where accurate measurements were given, were 2.5 by 1.3 cm., and the tumors and pulmonary changes reported in the present paper were representative of the group previously reported.

SUMMARY

Two cases of bronchial lipoma are reported which brings to 20 the total number of cases in which surgery was performed. Five additional cases have been recorded from autopsy material.

Lipomas as well as other benign bronchial neoplasms are capable of causing irreparable pulmonary parenchymal damage and on occasion may result in destruction of the host. It is for these reasons that early diagnosis and surgical treatment are of great importance.

Permission obtained from the AFIP, Capt. W. M. Silliphant, Director, for the incorporation of data from Case 2 is gratefully acknowledged.

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EFFECT OF EXPERIMENTAL PULMONARY VALVULAR INSUFFICIENCY ON THE CIRCULATION

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BARGER, Roe, and Richardson[1] produced pulmonary valvular insufficiency in 3 dogs by means of damage to 1 to 3 cusps. These workers concluded that pulmonary insufficiency alone had little effect on mean right auricular pressure or work tolerance in the dog, even after months of strenuous exercise. Ellison, Brown, Hague, and Hamilton[2] studied 10 dogs who survived total pulmonary valvectomy for 1 to 14 months. None of these dogs presented evidence of heart failure. Right ventricular end-diastolic pressures, cardiac outputs, and heart volumes determined by x-ray studies remained normal. In an earlier paper, Fowler, Mannix, and Noble[3] found no increase of right ventricular diastolic pressure in 8 dogs studied 2 weeks to 6 months after removal of one or portions of 2 pulmonary valve cusps.

The purpose of the present study was to evaluate the long-term effect in dogs of more extensive pulmonary valve resection than was done in our earlier work.[3] Since our findings differ in some respects from those of Barger and co-workers[1] and those of Ellison and associates,[2] it is believed worthwhile to present them.

MATERIALS AND METHODS

Attempted total pulmonary valvectomy was done in 18 mongrel dogs under anesthesia with pentobarbital sodium, 30 mg. per kilogram. Right ventricular circulation was interrupted for 2 minutes or less, by occlusion of the superior vena cava, inferior vena cava, and coronary sinus. After the right ventricle had expelled most of its blood, the main pulmonary artery was clamped. The right ventricle was opened, and the valve cusps were excised under direct vision. The edges of the right ventricular incision were approximated by a Potts clamp, and the circulation was re-established. The operative wound was then closed with interrupted silk sutures.

Right atrial pressures were recorded through a cardiac catheter with the chest closed. Right ventricular and pulmonary arterial pressures were recorded simultaneously preoperatively and postoperatively through No. 19 needles with chest and pericardium open. Pressures were detected by Statham P23D transducers or Sanborn electromanometers. Pressures were recorded upon the Sanborn Poly-Viso electrocardiograph or the Electronics for Medicine Research Recorder.

In 8 animals, surviving 11½ to 18 months, further studies were made. At 4 to 6 months after operation, right heart pressures were measured through a

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cardiac catheter, and cardiac outputs were measured by the direct Fick principle. At 11½ to 18 months after operation, the animals were sacrificed. Posteroanterior and right lateral teleroentgenograms of the chest were made preoperatively, at 4 to 6 months after operation, and prior to final sacrifice. Twelve lead electrocardiograms were made preoperatively, at 4 to 6 months after operation, and prior to final sacrifice.

Just prior to final sacrifice, other observations were made. Right atrial pressure and direct Fick cardiac outputs were determined in the anesthetized, closed chest animal. Electrical stimulation of the four extremities was then begun at a frequency of 284 per minute, with a pulse duration of 0.022 sec. and a current of 50 Ma. Stimulation was carried out for 10 minutes, with direct Fick cardiac output measurement during the last 3 minutes. After a 30-minute rest, the direct Fick cardiac output was again measured. Electrical stimulation and measurement of cardiac output were then repeated as before. The animal's chest was then opened under positive pressure respiration. Simultaneous measurement of right ventricular and pulmonary arterial pressure was made, and photographs of the heart were taken. The animal was then sacrificed. Liver biopsy was taken and the peritoneal cavity inspected for evidence of ascites. The heart was removed, weighed, and the pulmonary valve area was examined. The heart was then fixed in formalin for 2 to 4 weeks. Right and left ventricular weights were then determined by the method of Herrmann.[4]

RESULTS

Satisfactory pulmonary valvectomy was performed in 18 animals. Of these, 8 failed to survive the operation or immediate postoperative period because of excess blood loss, pneumothorax, or ventricular fibrillation associated with right ventricular dilation after valvectomy. One animal died one month postoperatively of extensive bronchopneumonia. The right ventricle and right atrium of the animal were dilated at autopsy. Another animal died 4 months after operation following a cineangiocardiogram, which demonstrated regurgitation of dye from pulmonary artery to right ventricle. Thus, 8 animals remained. These were sacrificed after 11½ to 18 months' observation, during which time they were allowed to exercise frequently and displayed no limitation of activity. Two animals had litters of 9 puppies each without apparent difficulty.

Autopsies were done in 10 animals surviving the operation. All 3 valve cusps had been removed in 5 animals; 2 cusps had been removed in 4; one cusp had been removed in one. Fig. 1 shows the pulmonary valve area of Dog 15B; only fragments of valve cusps are remaining. This animal was sacrificed 11 months and 3 weeks postoperatively.

At the time of the original operation, right ventricular and right atrial dilation occurred in each animal following pulmonary valvectomy. In 3 of the 18 animals, the right ventricle was dilated to an extent which rendered closure of the pericardium impossible. At the time of final thoracotomy prior to sacrifice, the right ventricle appeared dilated in each animal, but only after the pericardium was opened. Fig. 2 shows right ventricular dilation in a photograph of the heart of Dog 17B taken during thoracotomy just prior to sacrifice after survival of 11½ months. All 3 pulmonary cusps had been removed.

Teleroentgenograms of the chest, taken just prior to sacrifice, showed definite evidence of cardiac enlargement in only 2 animals, Dogs 14B and 20B. In each of these, the pericardium could not be closed at the time of the original operation because of right ventricular dilation. The anteroposterior and right lateral teleroentgenograms of Dog 20B are shown in Figs. 3 and 4. The preoperative films are on the left in each figure.

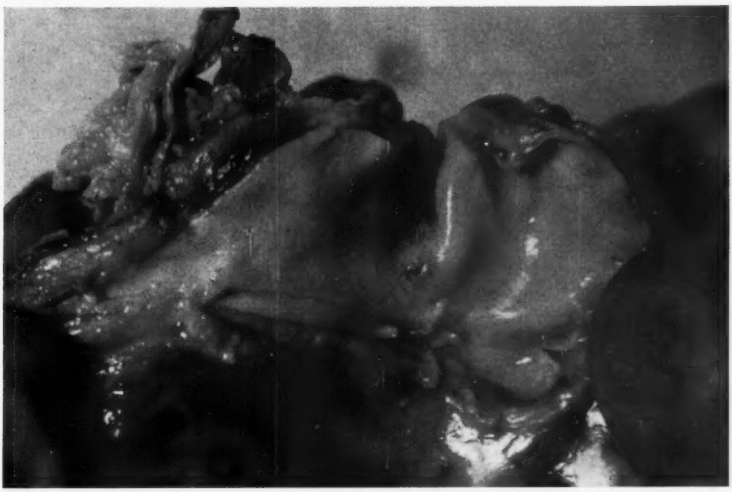

Fig. 1.—Photograph of pulmonary valve area of Dog 15B. This animal was sacrificed 11 months and 3 weeks postoperatively. Only fragments of the 3 pulmonary valve cusps are remaining.

Fig. 2.—Photograph of heart at thoracotomy of Dog 17B, 11½ months after removal of 3 pulmonary valve cusps. Note right ventricular dilation.

The results of pressure studies are given in Table I. Also given is the number of pulmonary valve cusps removed as established by autopsy. PA and RV pressures were obtained satisfactorily in 11 animals immediately before and

immediately following valvectomy. It may be seen that PA diastolic pressure fell in each instance; control values were 4 to 19 mm. Hg; a fall of 3 to 14 mm. Hg to values of 0 to 8 mm. Hg occurred following valvectomy. Right ventricular

Fig. 3.

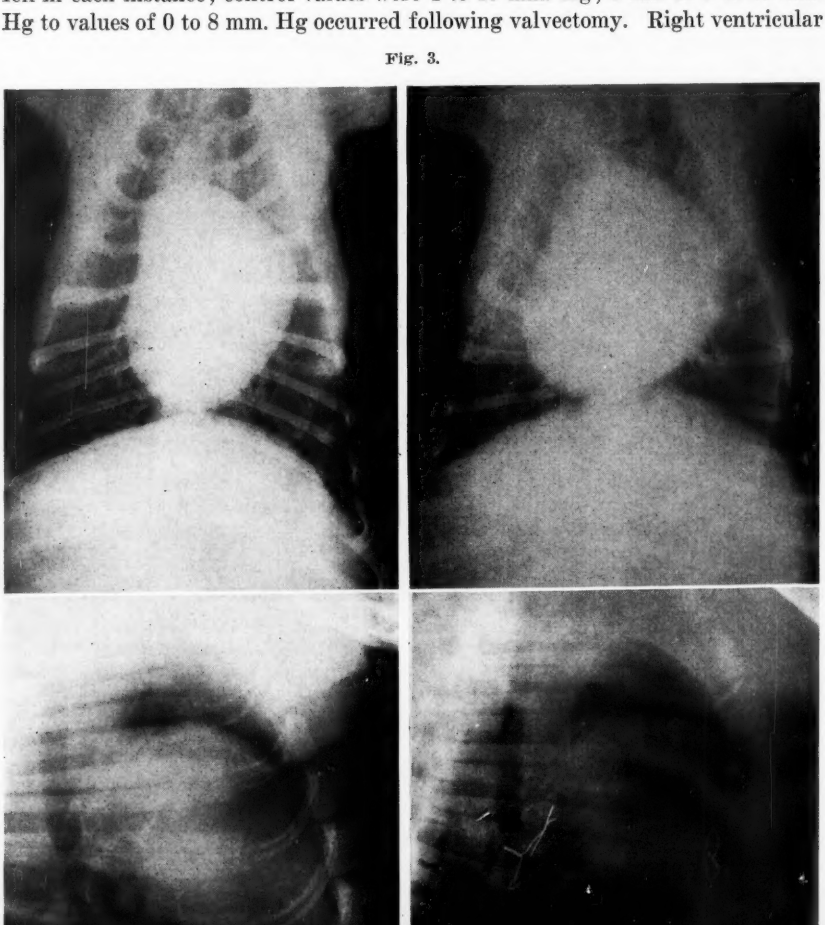

Fig. 4.

Fig. 3.—Anteroposterior teleroentgenograms of the chest in Dog 20B. On the left is reproduced the preoperative film. On the right is the roentgenogram taken 11½ months after removal of all 3 pulmonary valve cusps, showing increase in cardiac size. The pericardium could not be closed postoperatively in this dog.

Fig. 4.—Right lateral teleroentgenograms of Dog. 20B which shows increase in cardiac size 11½ months after total pulmonary valvectomy. The preoperative film is on the left.

diastolic pressure rose in seven instances. Where measured, right atrial pressure rose 0.5 to 1.5 mm. Hg immediately after operation. At the time of final study, one animal expired before pressures could be measured. At this time, right ventricular diastolic pressures were, in general, lower than immediately postoperatively. Fig. 5 shows simultaneous right ventricular and pulmonary arterial pressures of Dog 20B, showing RV pressure of 23/1.5 and PA pressure of 23/4 mm. Hg just prior to sacrifice, 11½ months after total pulmonary valvectomy. Right atrial pressures, however, were approximately as high or higher in three instances where comparisons could be made.

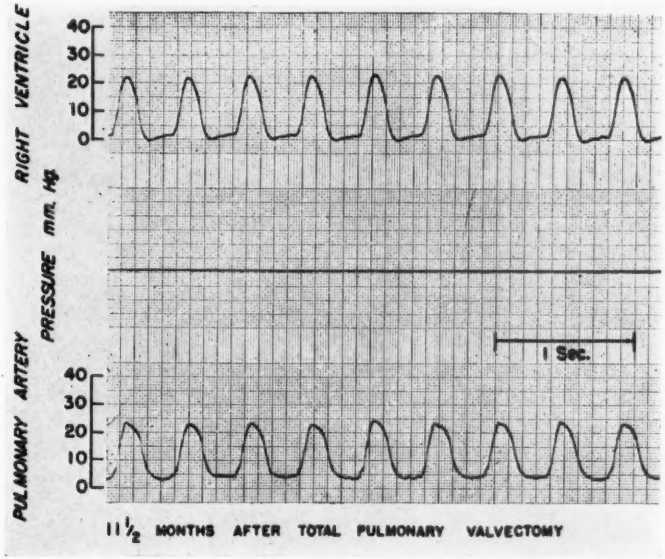

Fig. 5.—Simultaneous right ventricular and pulmonary arterial pressure record of Dog 20B. Pressures were taken 11½ months after total pulmonary valvectomy. RV pressure is 23/1.5 mm. Hg and PA pressure is 23/4 mm. Hg. PA pressure was 24/14 mm. Hg preoperatively.

Cardiac outputs were measured 4 to 6 months after operation in the survivors (Table II). The mean of cardiac outputs at this time was 199 ± 21 (S.E.) c.c./Kg./min. This value was compared with the mean of cardiac outputs in a control series of 52 normal anesthetized dogs studied in this laboratory, whose values were 161 ± 7.7 c.c./Kg./min. The difference was not significant statistically, $p > 0.05$. At the time of final study, the mean of cardiac outputs was 111 ± 14.9 c.c./Kg./min. This value was significantly lower than in the control group, $p < 0.05$, and was significantly lower than in the first postoperative study, $p < 0.05$. The first electrical stimulation increased mean cardiac output 178 ± 29 c.c./Kg./min., $p < 0.01$. Mean cardiac output fell to 131 c.c./Kg./min. 30 minutes after exercise and rose to 285 c.c./Kg./min. during electrical stimulation. The mean increase of 154 ± 33 c.c./Kg./min. was significant, $p < 0.01$.

TABLE I. RIGHT HEART PRESSURES, MM. HG, BEFORE AND AFTER PULMONARY VALVECTOMY

DOG NO.	CUSPS REMOVED	INITIAL STUDY						FINAL STUDY		
		BEFORE			AFTER					
		PA	RV	RA	PA	RV	RA	PA	RV	RA
1B	1	20/12	23/1.6		23/7	18/6		28/9	27/1.5	4.3
8B	2	9/4	13/1.2		22/1	16/1		25/4	20/0	7.0
9B	2	21/10	22/0		8/5	10/4		21/4.5	18/0	4.8
10B	–	18/11	18/2		16/1	23/0.5				
12B	3	21/11	19/2		23/0	36/1				
14B	2	27/19	31/0		26/5	25/2		20/5*	45/2.5*	3.8
15B	3	24/14	23/4	4.5	29/8	40/6	5.8	27/4	27/6	5.5
16B	3	20/13	20/4	3.5	30/5	28/4	4.5			
17B	3	14/7	15/2	2.8	25/4	26/4	3.8	25/6	20/1	3.8
18B	2	18/12	14/0.6	3.0	33/5	38/3	4.5			
20B	3	24/14	24/1	2.5	26/6	19/5	3.0	23/4	23/1.5	6.5

PA and RV pressures were measured with chest open.
RA pressures were measured with closed chest; 5 cm. up from table is zero for RA pressures.
*Catheter pressures.

TABLE II. CARDIAC OUTPUTS IN C.C./KG./MIN. IN 8 DOGS SURVIVING PULMONARY VALVECTOMY 11½ TO 18 MONTHS

DOG NO.	4 TO 6 MONTHS AFTER OPERATION	11½ TO 18 MONTHS AFTER OPERATION			
	REST	REST	1ST ELECTRICAL STIMULATION	2ND REST	2ND ELECTRICAL STIMULATION
1B	180	102	157	120	179
8B	117	67	344	83	396
9B	238	133	281	117	259
14B	302	52	299	136	198
15B	178	127	325	130	304
17B	249	138	348	195	392
18B	175	–	–	–	–
20B	153	160	274	137	265
Mean ± S.E.	199 ± 21	111 ± 14.9	290 ± 24.7	131 ± 11.7	285 ± 32.3

None of the sacrificed animals had evidence of ascites at autopsy. Appreciable weight gains (up to 5 Kg.) were recorded during the year period of observation, but these were presumably due to improved nutrition.

Twelve lead electrocardiograms taken preoperatively and at restudy in 5 animals showed no evidence of right ventricular hypertrophy.

The heart weight/body weight ratio was slightly above the upper normal of 0.00994 given by Herrmann[4] in 2 animals in which values of 0.0105 and 0.0100 were obtained; however, right ventricular/body weight ratios were each within the normal ranges of 0.00178 to 0.00400 given by Herrmann.

Liver biopsy showed slight hepatic cell degeneration and early fibrosis in the central vein areas in only 2 animals, Dogs 15B and 17B.

DISCUSSION

There are several indications in the study described above that severe pulmonary valvular insufficiency is not a benign lesion in its effect upon the circulation. Although our 8 animals which survived approximately one year were still apparently well, there were evidences of right heart dilation at the time of valvectomy and at the time of final sacrifice in each animal. In 3 of 10 surviving animals, right ventricular dilation at the time of valvectomy was so great as to preclude pericardial closure. In addition, the mean resting cardiac output, although normal 4 to 6 months postoperatively, was below normal approximately one year postoperatively. In this respect, our conclusions differ from those of Barger and co-workers,[1] and from those of Ellison and associates.[2] Also, we failed to find a systolic pressure gradient between right ventricle and pulmonary artery in the presence of pulmonary insufficiency as described by Ellison and co-workers.[2]

In man, primary pulmonary valve disease as a cause of pulmonary insufficiency is uncommon, and it is thus difficult to gain a clinical impression of the importance of this disturbance. McGuire and McNamara[5] described patients with organic pulmonary insufficiency; since other valves were involved in each of their patients, it is difficult to know what effect was produced by the pulmonary insufficiency. Kohout and Katz[6] reported a case in which a clinical diagnosis of pulmonary insufficiency was made. This patient showed evidence of right heart enlargement and had had heart failure. However, ventricular septal defect was probably present in addition to the lesion of the pulmonary valve. Campeau, Ruble, and Cooksey[7] reported a case of congenital absence of the pulmonary valve. Although their patient had congestive heart failure, it is impossible to ascertain the relative importance of the pulmonary valve lesion, since the patient had also a ventricular septal defect and a single coronary artery with myocardial infarction. Ford, Hellerstein, Wood, and Kelly[8] reported a case of pulmonary insufficiency associated with a congenital bicuspid pulmonary valve. Their patient, a woman of 43, developed right heart failure. She had also inactive pulmonary tuberculosis. These authors concluded from a review of the literature that congenital pulmonary incompetence due to bicuspid valve does not produce congestive heart failure unless other cardiac or pulmonary disease is present. Kjellberg, Mannheimer, Rudhe, and Jonsson[9] described a boy of 7 years in whom a clinical diagnosis of isolated pulmonary valvular insufficiency was made. This patient showed no evidence of right ventricular enlargement.

The effect of pulmonary valvular insufficiency upon the circulation is of importance, however, because of its occurrence as a complication of incision or partial excision of the congenitally stenotic pulmonary valve. Blount, McCord, Mueller, and Swan[10] stated that pulmonary insufficiency complicating the operation for pulmonary stenosis in 5 patients had produced no significant hemodynamic alterations, although they noted an increase in heart size postoperatively and thought that further observation was necessary. However, we have recently observed the development of right heart dilation in a boy of 6 years,

who had clinically and by right heart catheterization, only infundibular and valvular pulmonary stenosis. Following operation, which consisted of partial infundibular resection and incision of the pulmonary valve, the patient developed the murmur of pulmonary insufficiency and roentgen evidence of right heart dilation and abnormal peripheral pulmonary arterial pulsation. This was in evidence 3 months postoperatively. Cardiac enlargement of the same degree was still present 8 months postoperatively. One cannot be certain that this patient did not have some additional lesion, such as a small ventricular septal defect.

SUMMARY

Severe pulmonary valvular insufficiency was produced in 18 dogs, each of which developed right ventricular dilation postoperatively. In 3 animals, right ventricular dilation was so extensive that the pericardium could not be closed. Ten dogs survived the immediate postoperative period, and 8 dogs survived 11½ to 18 months and were sacrificed. There was no evidence of right heart failure nor of limitation of activity in the surviving dogs. Pulmonary artery and right ventricular pressure records showed evidence of severe pulmonary insufficiency. The mean of direct Fick cardiac outputs was normal 4 to 6 months after valvectomy and was below normal 11½ to 18 months after operation, but could still be increased by electrical stimulation of the extremities. At thoracotomy, after opening the pericardium, the right ventricle appeared dilated in each animal 11½ to 18 months postoperatively. Two animals of 8 long-term survivors showed evidence of cardiac dilation on teleroentgenograms. Two animals showed evidence of hepatic congestion on liver biopsy 11½ to 18 months after operation. It is concluded that severe pulmonary insufficiency in the dog is not innocuous even as an isolated lesion.

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PULMONARY FUNCTION BEFORE AND AFTER PULMONARY RESECTION IN TUBERCULOUS PATIENTS

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NUMEROUS reports in the medical literature indicate the interest that has centered on the reduction of pulmonary function accompanying resection of residual pulmonary foci following medical therapy of tuberculosis. Few studies have been reported, however, of groups of patients who had undergone resection without some concomitant space-filling procedure.

Loss of pulmonary function in patients with chronic pulmonary disease has been considered proportional to the extent of involvement of one or both lungs. Clinical and roentgenographic studies have not always provided a reliable index of the functional pulmonary capacity.[1, 2] Consequently, most studies of the effect of disease and of various types of therapy on pulmonary function have included preoperative and postoperative spirometric methods.[3-22] Bronchospirometry has also been employed when available and when unilateral variations have been anticipated.

There has been some controversy regarding the relation of loss of function to the number of segments resected. Van der Drift[4] and Limburg[5] indicated that pulmonary function is reduced less after segmental resection without complications than after lobectomy in patients with tuberculosis. They further stated that in larger unselected groups the reduction of function was similar whether segmental resection or lobectomy was carried out. Limburg suggested that this lack of difference might be caused by the greater incidence of complications among patients having segmental pulmonary resection. Taylor and associates[3] stated that loss of function following uncomplicated operations in a group of men with tuberculosis appeared to be unrelated to the increasing volume or number of segments resected, if less than a pneumonectomy. This, they thought, suggested the predominant influence of more constant factors.

However, Smith and associates,[7] in studying a group of young adults, mostly men, with bronchiectasis, stated that operation was found to produce further ventilatory loss proportional to the extent of the resection. They also

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reported that a relatively uniform ventilatory loss which followed excision of little or no pulmonary tissue in 11 tuberculous men was related to a "thoracotomy effect" or change in the thoracic wall.

Several authors[4, 5, 8, 12] have pointed out that the amount of functioning lung removed determines the degree of impairment of function, and have stressed the importance of preserving as much normally functioning pulmonary parenchyma as possible in various chronic conditions.

The study reported here was undertaken to provide additional evidence on the effects of unilateral resection for tuberculosis on pulmonary function.

MATERIAL AND METHODS

Twelve men and 11 women who were patients at the Mineral Springs Sanatorium, Cannon Falls, Minnesota, were studied. Their ages varied from 21 to 62 years (average 39 years). Preoperative therapy for pulmonary tuberculosis consisted of modified rest in bed and chemotherapy with two or more drugs for an average period of 8 months. Resection was limited to bronchopulmonary segments containing residual areas of disease considered to be a threat to the patient's future. Segmental resection was done in preference to lobectomy if salvageable segments were found in the diseased lobes. No patient in this study had less than one segment removed. If a wedge resection of a small nodule was done in addition to removal of one or more segments, this was not counted in classifying a patient according to the number of segments removed. No space-filling procedures were done. Each patient's progress was followed by clinical examination, cultures of sputum or fasting gastric contents, and roentgenographic studies, including tomograms. Only patients without postoperative complications were included in this study. Patients who required somewhat prolonged pleural suction for pulmonary re-expansion, bronchoscopy for aspiration of temporarily retained secretions, or thoracentesis for effusion were not considered to have significant complications.

Pulmonary function studies were performed at the Mayo Clinic, usually within 2 weeks before operation. They were repeated 2 to 6 months after operation, usually 1 or 2 weeks prior to dismissal from the Sanatorium. Spirograms and total lung volume and its subdivisions were determined with a spirometer of Benedict-Roth type and the open circuit nitrogen elimination method for residual volume[23] modified as follows: Oxygen was inspired from one Neoprene bag of about 150 L. capacity, and expired gas was collected in another. Both bags were enclosed in a box, also connected to a monitor spirometer providing automatic correction for "switch-in" error and a continuous recording of respiratory excursions. The nitrogen concentration of gases near the mouthpiece was recorded continuously with a nitrogen meter and a photokymograph both to aid in detection of inspiratory leaks and to record the maximal concentration of nitrogen of the alveolar samples delivered after 7 minutes of oxygen inhalation, hereafter called the "nitrogen washout index." Normal values for lung volumes were estimated with the formula of Bateman[24] and for maximal breathing capacity with that of Baldwin and associates.[25] Arterial oxygen

saturation was determined by a recording "double-scale" ear oximeter.[26] Bronchospirometry was performed with modified Carlen's catheters in the supine position in the usual manner[27] for 20 patients.

Control measurements of lung volumes and maximal breathing capacity were obtained on 10 healthy adults (5 men and 5 women), repeated after periods of 2 weeks to 4 months.

RESULTS

Preoperative Pulmonary Volumes and Maximal Breathing Capacity.—Table I and Fig. 1 present the values obtained on healthy subjects and the preoperative values for the patients, classified in relation to the number of segments ultimately resected. The ages in the groups of patients did not differ greatly.

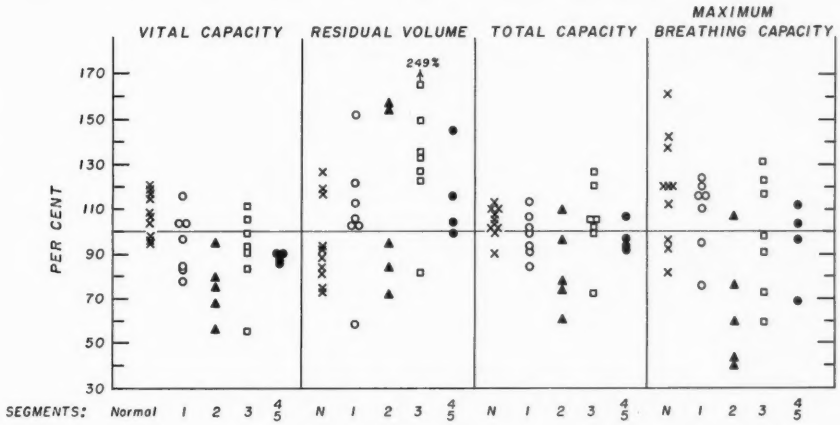

Fig. 1.—Preoperative values in tuberculous patients. Per cent of predicted normal.

TABLE I. AVERAGE VALUES OF PREOPERATIVE PULMONARY FUNCTION IN PER CENT OF PREDICTED NORMAL

SEGMENTS RESECTED LATER	SUBJECTS	MEAN AGE (YEARS)	VITAL CAPACITY	RESIDUAL VOLUME	TOTAL CAPACITY	MAXIMAL BREATHING CAPACITY
0 (Normals)	10	27	108	95	105	119
1	7	31	95	108	98	109
2*	5	43	74	112	83	63
3	7	43	91	142	105	99
4-5	4	41	90	116	98	95

*Low values attributed to significant bilateral disease in 3 of 5 patients.

The pulmonary volumes observed in the normal subjects agreed satisfactorily with predicted values, although the estimated maximal breathing capacities were usually somewhat smaller than observed.

Within each of the four groups of patients, there was considerable variation in the values for lung volumes and maximal breathing capacity, but the mean values were near normal except in the "two-segment" group, which had lower average values for vital, total, and maximal breathing capacities than predicted.

This was caused by low values obtained on 3 of the 5 subjects who had had severe contralateral disease treated many years previously by pneumothorax.

Although the contralateral lungs in the 3 patients showed only minor roentgenographic abnormalities at the time of operation, it was apparent that contralateral disease had been extensive on the basis of the history, the previous thoracic roentgenograms, and preoperative bronchospirometric data which showed that the lung to be operated on was contributing a greater portion of the total function than the contralateral lung. The one patient in the three-segment group with markedly reduced values of vital, total, and maximal breathing capacity had undergone a contralateral five-rib thoracoplasty 6 years previously.

The size of the residual volume was related to age. Increased residual volumes, amounting to more than 125 per cent of the absolute value predicted for young adults, and also to more than 35 per cent of the total capacity, were found in 4 of the 6 patients more than 50 years of age and in only 2 of 17 patients less than 50 years of age.

The arterial oxygen saturation was normal (95 to 98 per cent) in all patients tested, both at rest (20 patients) and during several minutes of step-test exercise (12 patients). In only 4 of 23 patients was the pulmonary nitrogen washout index abnormal (>2.5 per cent), indicating the presence of retarded alveolar ventilation of some part of the lungs. These 4 men did not show the absolute and relative increases of residual volume that are usually associated with abnormal nitrogen indexes in chronic diffuse obstructive emphysema.

Postoperative Pulmonary Volumes and Maximal Breathing Capacity.—Table II and Fig. 2 show that, in control normal subjects, average pulmonary volumes were unchanged within 2 weeks to 4 months. Vital capacity and total capacity varied only within 94 to 107 per cent of the initial test.

In the patients, the postoperative value for vital capacity was less than the preoperative value in 22 of 23 cases, and the average reduction was similar in all 4 groups. In contrast, the residual volume and functional residual capacity were not consistently changed in patients having one or two segments removed, but were reduced in 9 of 10 patients in the three and four to five segment groups.

The total capacity was reduced in all groups, but not in a close relation to the number of segments resected. Postoperative values for maximal breathing capacity varied considerably from the preoperative value in many patients, but in no case were they reduced more than 30 per cent. In all groups, the average preoperative and postoperative values were similar for maximal breathing capacity and for the percentage of the "timed" or rapid vital capacity expired in one second (average 80 per cent). The average value of maximal midexpiratory flow[28] was 2.2 L. per second before and 1.9 L. per second after operation.

Bronchospirometric values are shown in Table III and Fig. 3. In 18 of 20 cases, the percentage of the total vital capacity contributed by the lung operated on was reduced postoperatively. An attempt was made to determine whether the postoperative reduction of total vital capacity could be accounted for entirely by the reduction in the vital capacity of the lung that was operated

on. The sum of the vital capacities of the two lungs recorded during bronchospirometry in the supine position was somewhat less than the total vital capacity measured in the sitting position about an hour before bronchospirometry. Therefore, the vital capacity of each lung was multiplied by the ratio of standard vital capacity to total bronchospirometric vital capacity for each of the

TABLE II. POSTOPERATIVE PULMONARY FUNCTION
(AVERAGE EXPRESSED IN PER CENT OF PREOPERATIVE VALUES)

SEGMENTS INVOLVED	SUBJECTS	VITAL CAPACITY	RESIDUAL VOLUME	TOTAL CAPACITY	FUNCTIONAL RESIDUAL CAPACITY	MAXIMAL BREATHING CAPACITY
0*	10	100	101	100	100	115
1	7	89	106	94	101	105
2	5	81	92	86	92	134
3	7	88	79	84	88	92
4-5	4	83	80	83	81	95

*Tests repeated in 2 to 16 weeks on normal subjects.

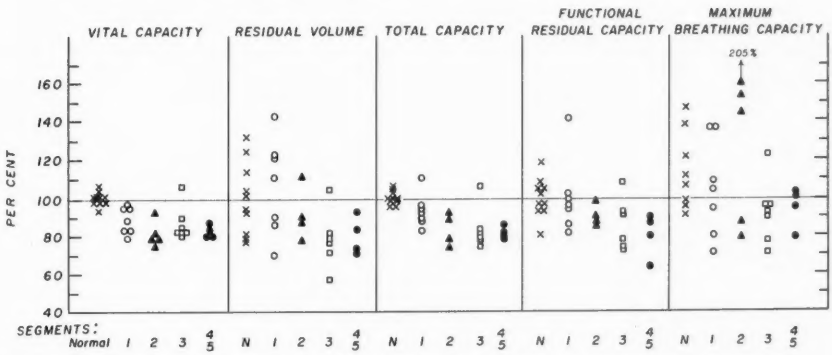

Fig. 2.—Postoperative values in tuberculous patients. Per cent of preoperative values.

preoperative and postoperative measurements. Thus, the postoperative unilateral reductions could be compared with the total reductions. In 13 of 20 patients, the unilateral loss was calculated to be between 75 per cent and 125 per cent of the total loss, indicating a reasonable accounting for the total loss. In 2 patients, the unilateral loss was more than 125 per cent of the total loss, indicating that the contralateral lung had increased its vital capacity. In 5 patients, the unilateral loss was less than 75 per cent of the total loss, indicating some contralateral impairment after operation.

With a few exceptions, reductions of oxygen uptake and respiratory minute volume of the lung operated on were found. The magnitude of reductions was not closely related to the number of segments removed. The amount of reduction in the percentage of the total minute volume contributed by individual lungs was more closely related to the amount of reduction of vital capacity than to that of oxygen uptake (Fig. 4).

OTHER STUDIES

Postoperatively, arterial oxygen saturation was normal in each of the 21 patients measured at rest and in the 15 patients measured during exercise. Of

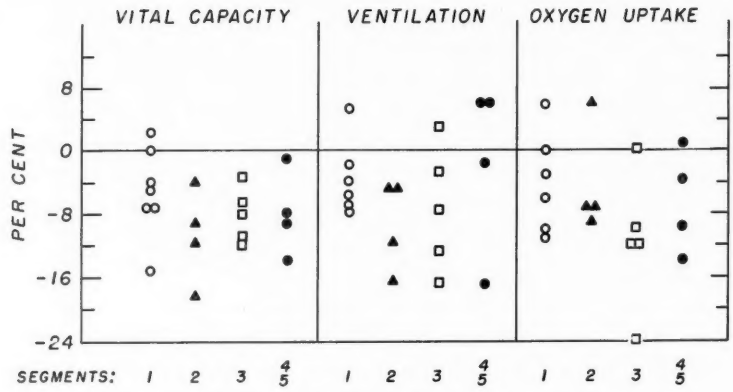

Fig. 3.—Change in lung operated on in per cent of total function.

TABLE III. AVERAGE BRONCHOSPIROMETRIC VALUES

NUMBER OF SEGMENTS REMOVED	SUBJECTS	PER CENT OF TOTAL FUNCTION PERFORMED BY THE LUNG OPERATED ON						RATIO OF POSTOPERATIVE TO PREOPERATIVE VALUES LISTED IN COLUMNS TO THE LEFT, PER CENT		
		VITAL CAPACITY		MINUTE VENTILATION		OXYGEN UPTAKE				
		PREOPERATIVE	POSTOPERATIVE	PREOPERATIVE	POSTOPERATIVE	PREOPERATIVE	POSTOPERATIVE	VITAL CAPACITY	MINUTE VENTILATION	OXYGEN UPTAKE
1	7	44	40	43	40	41	37	88	91	90
2*	4	59	48	56	47	51	45	80	81	92
3	5	48	40	51	43	47	35	82	86	78
4-5	4	44	36	43	41	39	32	81	94	84

*High preoperative values attributed to prominent contralateral disease in 2 patients.

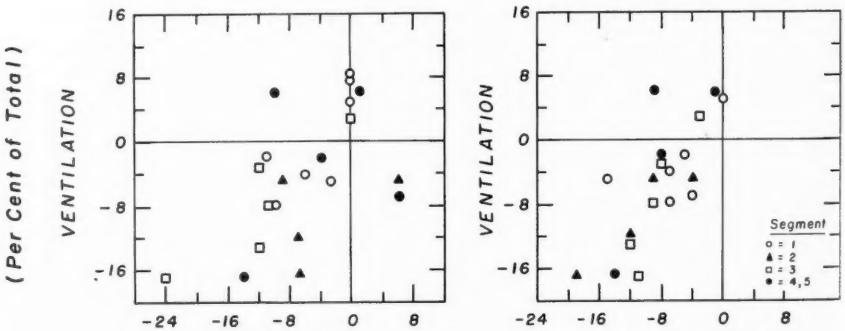

Fig. 4.—Relation of change in ventilation of the lung operated on to changes in oxygen uptake and vital capacity.

the 4 patients who had abnormal nitrogen washout indexes preoperatively, 2 remained abnormal, but 2 had normal indexes postoperatively, suggesting more uniform alveolar ventilation. After operation, 3 other patients had abnormal indexes. Estimates of the ratio of alveolar ventilation rate to functional residual capacity suggested that in one case the abnormal index probably indicated less uniform alveolar ventilation, and in another that the abnormality of alveolar ventilation had existed preoperatively, but had been masked by an increased rate of ventilation.

EFFECT OF SEX AND AGE

The postoperative changes described in preceding paragraphs were of similar magnitude in both sexes and in patients more than or less than 40 years of age.

MULTIPLE RESECTIONS

Three patients underwent a second contralateral resection of one or two segments and were again tested 2½ to 4½ months later. In all, the maximal breathing capacity was unchanged after the second operation, the lung volumes (vital capacity, and so forth) were unchanged in one patient, but were further reduced by 5 to 10 per cent in 2 patients. A reduced contribution by the lung undergoing the second operation was observed in all cases, amounting to 5 to 15 per cent of the total function. Arterial oxygen saturations were normal at rest and during exercise in all patients.

COMMENT

The changes noted after operation in this study were similar to those of other reports. On examination, several months after resection of only one segment, or even after local enucleation of tuberculomas,[7] reductions of about 10 per cent in vital capacity are observed consistently,[3, 4, 18, 21, 22] with similar reductions of total capacity.[7, 21, 22] Reductions of residual volume or functional residual capacity are small and less consistent.[7, 21] Maximal breathing capacity and the one-second percentage of the rapid vital capacity are essentially unchanged. Resection of one segment causes the percentage of total ventilation, oxygen uptake, and vital capacity contributed by the side operated on to decrease about 5 percentage points, although the reductions may occasionally be as great as 20 per cent.[3, 4] Our data and those of Hirdes[21, 22] indicate that the loss of vital capacity on the side operated on largely accounts for the loss of total vital capacity.

After resection of two to five segments, the reduction in vital capacity,[3, 4, 18] residual volume, and total capacity[21, 22] is not much greater than after resection of one segment. The absence of a clear relationship between the number of segments removed and the volumetric reductions has been stressed by most authors. However, a trend of this type is apparent in our results and those of others.[3, 21]

Whereas the maximal breathing capacity is not consistently altered after resection of one segment, it is usually reduced 10 to 15 per cent after resection of four to five segments.[3, 4, 19, 21]

The reduction of oxygen uptake by the lung operated on is usually greater than the reduction of its minute volume. There is no close relationship between the reduction of function of the lung operated on (see Fig. 2) and the number of segments (up to four or five) removed.[3, 21] Although the average reduction of oxygen uptake by the lung operated on was found to be greater after lobectomy than after segmental resection by van der Drift,[4] this was usual only with the left upper lobe.

The arterial oxygen saturation was found to be usually normal after lobectomy or lesser resections for bronchiectasis and other lesions[7, 20] and was normal in all of our patients at rest or during exercise or both.

This and similar studies have attempted to determine the extent by which surgical procedures further alter the function of a diseased lung. The preoperative impairment shown by tests of both lungs and of the lung to be operated on varied markedly between patients and was not closely related to the number of segments ultimately removed. This was not surprising. The resections were performed to prevent recurrence of active tuberculosis by removing the danger of reactivation of a residual area of disease following chemotherapy. Very likely, previous involvement of portions of the lungs other than the circumscribed area removed had left its mark in functional impairment. This more diffuse involvement with healing and varying degrees of fibrosis would not necessarily be related to the number of segments containing caseous residues. Only that amount of pulmonary tissue necessary to remove significant residue was resected, as much grossly normal tissue as possible being preserved. Thus the surgeon would hope ideally to remove little functioning pulmonary tissue. If such were possible, and barring major complications, there might be little functional impairment following operation, and the functional status, relative to the preoperative conditions, should be rather uniform despite a varied number of segments removed. There might also be a rather uniform extent of alteration caused by changes in the thoracic wall and pleura. Such changes have been given the term of "thoracotomy effect."[3, 7]

It seems clear that the magnitude of reduction of pulmonary volumes is not greatly different after resection of one or several segments for tuberculosis. This suggests that a minimal, rather uniform, amount of functioning lung is in fact removed. In addition, some possible variability related to the number of segments removed is probably masked by the "thoracotomy" effect described as following enucleation of small nodules. However, there is a trend toward increasing reduction of pulmonary volumes that indicates that some sacrifice of ventilated lung is made. This trend is likely to be followed when a patient undergoes an additional resection.

Reduction in ventilatory capacity after resection for bronchiectasis was found by Smith and associates[7] to be proportional to the number of resected segments. That such a relationship is less evident after resection for tuberculosis than after resection for bronchiectasis may perhaps be explained by the fact that resection for bronchiectasis is directed at diseased bronchi. Parenchyma distal to some of the diseased bronchi might be contributing significant amounts to total function of the lung. Resection of these bronchopulmonary segments

should lead to reduction in function, depending on the number removed. By further contrast, our older tuberculous patients had as good functional result as the younger, whereas greater impairment has been reported after resections for bronchiectasis in older patients.[7]

There has been much concern over whether the increased expansion of the remaining pulmonary tissue that occurs after large resections represents or may lead to emphysematous changes. The reduction of functional residual capacity by about 20 per cent of the preoperative values after resection of four or five of the eighteen bronchopulmonary segments indicates the absence of an average increase of expansion of all remaining segments. However, this does not preclude a relatively greater expansion of the segments remaining on the side operated on than on the other. This condition would seem to be better designated as "compensatory overexpansion" than as "compensatory emphysema." Although after operation a minority of our patients had small increases of the residual volume/total capacity ratio, we do not feel justified in considering this as evidence of obstructive emphysema in the absence of obvious abnormalities of the distribution of alveolar ventilation, as indicated by the nitrogen washout index, or of evidence of expiratory obstruction, or of significant increases of absolute residual volume.

Despite the recognized limitations of the tests used to assess pulmonary function, it seems probable that, when the remaining lung is not deficient, the reductions of up to 20 per cent of various lung volumes that follow resection of one to five segments for pulmonary tuberculosis cannot be presently considered to represent a clinically significant impairment.

SUMMARY

To gain further knowledge of the effects of pulmonary resection, pulmonary function studies were performed on 23 adult tuberculous patients about 2 weeks before and 2 to 6 months after resection. There were 12 men and 11 women, with a mean age of 39 years. One to five bronchopulmonary segments were removed without additional space-filling procedures, following a period of combined chemotherapy and modified rest in bed averaging 8 months.

Postoperative changes varied within each of the groups classified according to the number of segments removed. The vital capacity was reduced in almost all cases, the average reduction being similar in all groups. In contrast, the average residual volume was unchanged in patients having one or two segments removed but was reduced in those having three to five segments removed. The total capacity was reduced in all groups but not in close relation to the number of segments removed. In all groups the average maximal breathing capacity and the per cent of rapid vital capacity expired in one second were unchanged postoperatively. The 7-minute alveolar nitrogen (breathing oxygen) was within or near normal limits in most cases before and after operation. The arterial oxygen saturation was normal in each case at rest and with mild exercises preoperatively and postoperatively.

Bronchospirometry, performed in 20 patients, showed that the over-all loss of function could be explained in most instances by unilateral changes.

Changes were similar in subjects more than or less than 40 years of age and in both sexes.

The reduction of pulmonary function is not greatly different after resection of one or several tuberculous pulmonary segments, suggesting that a minimal, rather uniform, amount of functioning lung is removed with the diseased tissue. Some possible variability related to the number of segments removed is probably masked by the "thoracotomy effect" described as following enucleation of small pulmonary nodules. However, there is a trend toward increasing reduction of pulmonary volumes, indicating that some sacrifice of ventilated lung is made. This trend is likely to be followed when a patient undergoes an additional resection.

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CONGENITAL COMMUNICATION BETWEEN THE RIGHT CORONARY ARTERY AND THE RIGHT ATRIUM

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LESIONS which cause continuous precordial murmurs that are to be distinguished from patent ductus arteriosus have had considerable coverage in the literature.[1, 2] Particularly is this true of aorticopulmonary septal defect,[3-12] ruptured aneurysm of an aortic sinus,[13, 14] and ventricular septal defect associated with aortic insufficiency.[15-19] Less emphasis has been made of yet other conditions such as arteriovenous communications, involving vessels of the thoracic cage,[20, 21] the subclavian[22, 23] or the internal mammary vessels,[24] and communications between the coronary arterial system on one hand and the coronary veins, a cardiac chamber, or the pulmonary trunk, on the other.

Herein is reported the case of an adult patient who had a communication between the right coronary artery and the right atrium. The cardiac failure exhibited is considered to have been caused by the arteriovenous shunt which accompanied the abnormal congenital vascular connection.

It is pertinent to emphasize, by the report of this case, that the coronary arterial system may be involved in congenital conditions which are, or which function, essentially like arteriovenous fistulas. Recognition of the condition in a given patient may lead the way to surgical obliteration of the abnormal communication. Such goals have been accomplished in isolated instances.[25-27]

CASE REPORT

Pertinent Clinical Features.—A woman, first noted to have congestive heart failure at the age of 48 years, was given appropriate treatment for this condition during the ensuing 5 years. At the end of this time, however, she died of intractable failure of the heart. Her history was noncontributory. She had not known that she had heart disease until she consulted a physician for complaints related to the heart failure.

The pertinent facts of her physical examination follow: The heart was greatly enlarged to the left. The point of maximal impulse was in the left anterior axillary line at the fifth and sixth intercostal spaces.

A loud, harsh, blowing, continuous precordial murmur was present. It was heard best anteriorly in the right fifth intercostal space. It also was loud over the lower sternal region. There was enlargement of the cervical veins and of the liver. On occasion, pulsation was noted in the latter organ.

On all examinations the patient was normotensive. A representative blood pressure reading was 110 mm. Hg systolic and 70 mm. diastolic. Usually, the rhythm was regular. Occasionally, ventricular extrasystoles were noted.

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Fig. 1.—Roentgenogram of thorax.

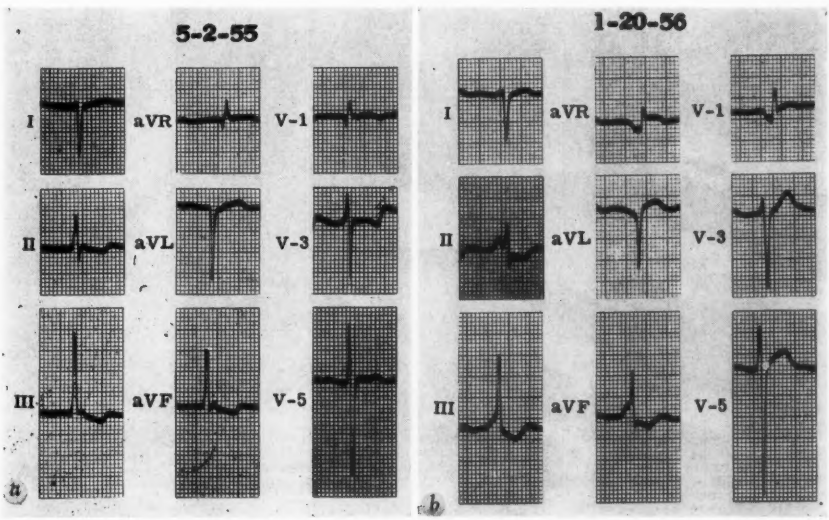


Fig. 2.—*a*, Electrocardiogram made on May 2, 1955. *b*, Electrocardiogram made on Jan. 20, 1956.

A roentgenogram of the thorax (Fig. 1) revealed evidence of cardiac enlargement, rounding of the right margin, and prominence of the pulmonary trunk. The electrocardiogram was difficult to interpret because of the probability of an anomalous excitation of the ventricles of the Wolff-Parkinson-White type (Fig. 2, *a* and *b*). Evident abnormalities were a right axis deviation, an unusual qR pattern in Lead V_1, and a nondiagnostic biphasic QRS (RS) in the left precordial leads. The degree of change in the ST segment in Leads aV_L and aV_F gave strong indication of ischemic injury of the anterolateral wall of the left ventricle.

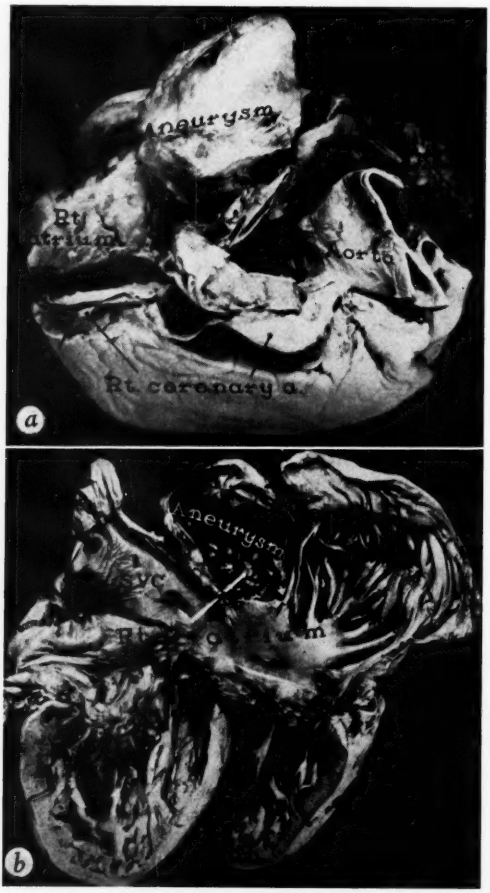


Fig. 3.—*a*, The base of heart viewed from above. A greatly dilated right coronary artery originates from the aorta. In the right atrioventricular sulcus the artery divides. A dilated branch proceeds posteriorly toward the right atrium and has an aneurysm on it in relation to the right atrium. The other branch continues in the right atrioventricular sulcus as the right coronary artery.

b, Interior of right side of heart. Shown is one of the communications which connects the right atrium with the aneurysmal part of the dilated branch of the right coronary artery (probe). The junction of superior vena cava and right atrium is indicated as *SVC*.

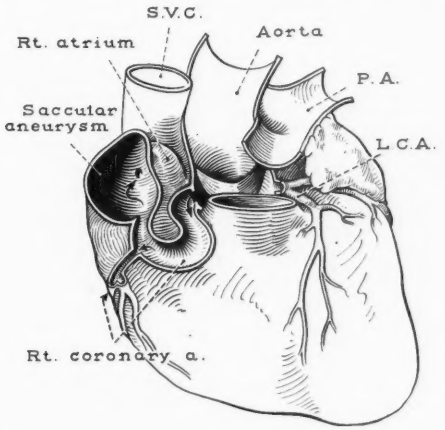
Pathologic Features.—At necropsy, significant findings were found in relation to the heart which showed anomalous communication of the right coronary artery with the right atrial cavity.

The heart was enlarged, weighing 500 grams. The enlargement was contributed mainly by hypertrophy of each ventricle. In its anterior wall, the right ventricular myocardium measured 0.5 c.c. in thickness; in its posterior wall it measured 0.8 c.c. The left ventricular wall measured 1.7 c.c. in thickness.

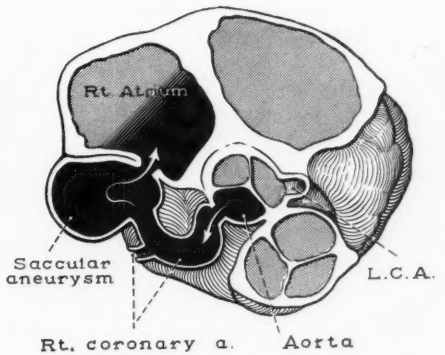
The sites of origin of the coronary arteries from the aorta were normal.

The right coronary arterial ostium was wide, measuring 6 mm. Immediately on exit from the aortic wall, the right coronary artery widened to a diameter of about 1.5 c.c.

The proximal part of this artery was tortuous to a point about 3 c.c. from the aortic wall. Here the right coronary artery divided into two branches (Figs. 3 and 4). One



a.



b.

Fig. 4.—*a*, In the case reported, the dilated coronary artery originated from the aorta and then divided into a wide branch which in turn became aneurysmally dilated and communicated with the right atrium. The other branch continued as the right coronary artery.

b, The perspective essentially equivalent to that shown in Fig. 3, *a*, wherein a branch of the right coronary artery communicates with the right atrial cavity.

branch, 4 mm. in diameter, was essentially a continuation of the right coronary artery in the right atrioventricular sulcus. The other branch was directed posteriorly toward the anterior wall of the base of the right atrial auricular appendage. It was tortuous and had essentially the same appearance as the proximal part of the parent right coronary artery.

As this branch approached the anterior wall of the right atrium, it dilated suddenly into a saccular aneurysm measuring about 3.5 by 5 cm. Whereas the tortuous portions of the

right coronary arterial system were thin, the saccular aneurysm had a tough gray fibrous wall containing calcium. Through its posterior wall, the saccular aneurysm communicated with the base of the right auricular appendage by way of three small openings, the largest of which was 3 mm. in diameter.

After giving off the dilated right atrial branch as described, the right coronary artery continued in a normal course and terminated as the posterior descending coronary artery. Just before entering the posterior interventricular sulcus the right coronary artery gave off a branch to the posterior lateral aspect of the left ventricle.

As is often found normally, an accessory artery distributed to the anterior wall of the outflow tract arose from the aorta just anterior to the origin of the main right coronary artery.

The left coronary artery measured 3 mm. at its origin, was devoid of disease, and terminated normally into the anterior descending and left circumflex branches. The terminal branches of the left circumflex coronary artery were distributed to the lateral wall of the left ventricle. Gross connections were not identified between the right and left coronary arteries on gross examination.

Histologic examination of a section taken through the dome of the aneurysm involving the branch of the right coronary artery revealed a small segment of arterial structure which was interrupted and then became continuous with the wall of the aneurysm (Fig. 5, *a* and *b*). The aneurysmal wall was composed of collagenous connective tissue and did not have identifiable arterial elements. The lining of the aneurysm was greatly thickened by the presence of many calcified amorphous masses. These masses had the appearance of calcified thrombotic material. No foam cells or cholesterol clefts were present.

A section taken through the dilated portion of the right coronary artery showed the media to be greatly thickened with muscle (Fig. 5, *c*). The elastic laminae were considerably disrupted and were represented only by occasional strands. The lining of the vessel in this portion was somewhat thickened by nonspecific fibrous tissue. The right coronary artery beyond its dilated portion and the anterior descending coronary artery did not show any remarkable features histologically.

The myocardium in the lateral portion of the left ventricle contained many wide spaces filled with blood (Fig. 5, *d*). Some had walls composed of connective tissue lined by layers of collagen and elastic tissue. These vessels appeared to be dilated sinusoids and some communicated with the left ventricular cavity. Other wide vessels in this region were obviously arteries.

COMMENT

In this case, the communication of a branch of the right coronary artery with the right atrium is representative of a condition described in a number of cases in the literature in which a coronary artery communicates with the right atrium, the right ventricle, or a coronary vein.[28-38]

At operation, Davis, Dillon, Fell, and Gasul[26] observed a branch of the left coronary artery communicating with the right ventricular cavity. Their 19-year-old patient had had a murmur that was suggestive of patent ductus arteriosus. Cardiac catheterization revealed arterialization of blood in the right ventricle. The anomalous communication was interrupted successfully.

In yet another case which was observed by Gasul, Fell, and Dillon,[39] a 9-month-old child had a continuous systolic and diastolic murmur heard best over the fourth and fifth intercostal spaces between the apex and the left sternal border. Because of the unusual location of the murmur it was considered that the child might have an arteriovenous communication involving the coronary system. An angiocardiogram revealed marked enlargement of the

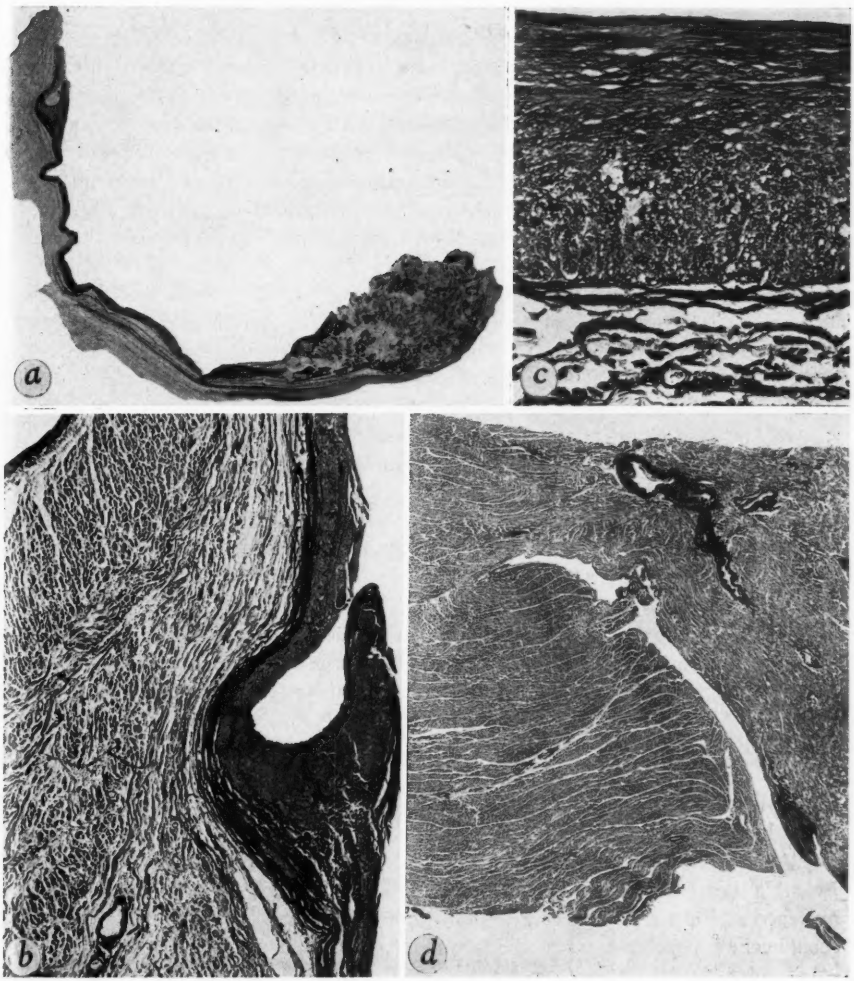


Fig. 5.—*a*, The aneurysm of the dilated branch of the right coronary artery. At one zone there is normal arterial structure which suddenly interrupts and beyond this there is nonspecific wall of the saccular aneurysm (see *b*, for details). Calcific material, which may be thrombotic in origin, is deposited on the inner surface of the aneurysm (Verhoeff's elastic tissue stain counterstained with van Gieson's connective tissue stain; ×2).

b, Higher magnification (×30) of junction of arterial wall and aneurysm illustrated in *a*.

c, Section through the dilated portion of the proximal segment of the right coronary artery shows evidence of medial hypertrophy. The internal and external elastic laminae are interrupted and represented only by fragments of elastic tissue. There is mild nonspecific fibrous thickening of the intima (Verhoeff's elastic tissue stain, van Gieson's connective tissue stain; ×100).

d, Lateral wall of the left ventricle. Within the myocardium are dilated spaces, some of which are thick-walled sinusoids; others are arteries. A relatively wide sinusoid communicates with the left ventricle shown in the lowermost portion of the illustration (Verhoeff's elastic tissue stain and van Gieson's connective tissue stain; ×5).

right coronary artery which appeared to end in a large dilated opacified region close to the cardiac apex. Cardiac catheterization revealed normal intracardiac pressures and evidence of a small left-to-right shunt at right ventricular level. At operation, a greatly dilated right coronary artery was found to enter the diaphragmatic portion of the right ventricle near the apex. The vessel was interrupted and the patient's course subsequently was uneventful.

In an adult patient with a continuous precordial murmur, Morrow[40] observed, on retrograde aortography, a communication between the aorta and the right atrium. At operation an anomalous communication between the right coronary artery and the right atrium was identified. The communication was closed at its right atrial aspect.

In the study of a human embryo, Essenberg[41] found that a branch of the left coronary artery penetrated the wall of the left atrium and then coursed through the cavity of the left atrium for a short distance and penetrated the right atrial wall. This vessel may have communicated with the right atrial cavity but the serial sections to determine this point were incomplete.

In the case of an adult patient, Garomella[42] observed that, at necropsy, a connection existed between an accessory branch of the left coronary artery and the right atrium.

Paul and associates[43] described the case of a 9-year-old boy with a continuous precordial murmur who, at operation, was found to have an enlarged right coronary artery that seemed to communicate with a tributary of the coronary sinus.

A common finding in the group of cases mentioned was the presence of a left-to-right shunt from a coronary artery into some point in the lesser circulation. Such a communication may give rise to one or more of the several complications of arteriovenous fistula anywhere in the systemic circulation. If the opening between the coronary artery and the right-sided chamber is sufficiently wide to cause a large shunt, the latter may be responsible for cardiac failure. The dynamics of arteriovenous fistula are such that bacterial endarteritis may occur in the abnormal channel.[36, 38]

An additional complication of the arteriovenous communication is that of ischemia beyond the point of the abnormal communication. The resulting myocardial ischemia may give rise to a picture which may simulate coronary insufficiency resulting from the common type of coronary disease, namely, atherosclerosis.

The abnormal vibrations generated by the arteriovenous shunt have frequently been described as the cause of a continuous thrill or of a to-and-fro murmur associated with a continuous thrill. Although in most instances wherein such a murmur was described, the location of the murmur was farther to the right and lower than that of typical patent ductus arteriosus, the latter condition has been suspected in some patients, as in the case of Davis and associates.[26] In some cases, such as the case of Espino Vela and associates,[38] the position of the murmur has suggested another type of arteriovenous communication, namely, an aneurysm of an aortic sinus (Valsalva) with rupture into a right-sided cardiac chamber.

The exact anatomic diagnosis of an arteriovenous communication, wherein a coronary artery takes part in the abnormal flow of blood, is not necessarily simple to make clinically. The classic findings at cardiac catheterization would be expected to give rise to evidence of an increased amount of oxygen in the blood in the chamber receiving the abnormal shunt. In the case of Davison and associates,[32] wherein the branch of the left circumflex coronary artery communicated with the coronary sinus, cardiac catheterization revealed arterialization of the blood in the right atrium. Such a finding coupled with abnormal murmurs of the type usually found in this condition also might be associated with a ruptured aneurysm of an aortic sinus. Retrograde aortography probably is a more exact method than cardiac catheterization for diagnosing the exact type of communication.[40]

Cases with the foregoing types of communications have basic functional features in common with connections between a main-stem coronary artery and the pulmonary trunk.[44-49] Moreover, in the condition in which one coronary artery arises from the pulmonary trunk while the other arises from the aorta, present-day evidence[50-52] supports the old concept of Brooks[45]: that an arrangement functionally similar to that in arteriovenous communications also exists.

Abnormal communication of a coronary artery with a right-sided cardiac chamber is expected to yield evidence of arterialization in the right side of the heart; less commonly a coronary artery communicates with a left-sided cavity. This results in findings that might be confused with aortic insufficiency. In the case of Lovitt and Lutz,[53] a branch of the anterior descending coronary artery communicated with the left ventricular cavity.

In Mozen's case,[27] an abnormal communication between the left coronary arterial system and the left atrium was recognized at operation. Interruption of the anomalous channel led to disappearance of associated murmurs and thrill.

An addendum to a paper by Paul and associates[43] presents a surgical experience of Dr. Robert E. Gross with a 16-year-old boy in whom a clinical diagnosis of patent ductus arteriosus and healed bacterial endarteritis had been made. At operation, a portion of the lateral wall of the left ventricle was found to be thin, bulging, and associated with a thrill. Though the situation of the major coronary arteries leading to this area was not ascertained, a postoperative diagnosis of "arteriovenous aneurysm, probably of the coronary system" was made.

Reid[54] described a gross communication between the apical portion of the left ventricle and the anterior descending coronary artery in the heart of an ox. A single opening is often described at the site of an anomalous connection in human beings, but at times there are several, indeed multiple, openings. Wilson and Grant[55] found a communication between the right coronary artery and the single ventricle of a malformed heart. In this case there was an aneurysm of the artery. Histologic examination revealed that the aneurysm opened into many blood-filled spaces within the myocardium. These spaces, in turn, opened into the ventricular cavity.

The association of a saccular aneurysm with an abnormal communication between the coronary artery and the cardiac chamber is frequently described and, moreover, in many reports the aneurysm is given a major position in the title and discussion. The aneurysm, however, is to be viewed simply as a complication of an arteriovenous communication. In all cases wherein such communications have been described, the artery proximal to the communication is greatly dilated as it was in the case reported herein. Associated with the right coronary arterial dilatation in our case was hypertrophy of the media of the artery proximal to the anomalous communication, a feature which is frequent in arteries proximal to arteriovenous communications anywhere in the body. Of interest with regard to genesis of the saccular aneurysm is the fact that associated with the dilatation there was interruption of the elastic layers of the dilated segment of artery. Such interruptions in the elastic layers probably represented the underlying cause for the development of the saccular aneurysm. In the case reported herein, a section of the saccular aneurysm revealed that for the most part it was composed of nonspecific connective tissue. At one point a segment of arterial wall ended abruptly and then communicated with the aneurysmal wall. It is probable, as suggested by Scott,[46] that the saccular aneurysm arose at the site of an arterial branch. The calcific material within the saccular aneurysm in the case reported is possibly derived from thrombotic material within the saccular aneurysm.

In one case the electrocardiogram gave suggestive evidence of ischemia in the anterolateral region of the left ventricle. This is of special interest in view of the fact that, histologically, the lateral wall of the left ventricle showed dilated arteries and sinusoids. This brings up for consideration the possibility that blood in the left ventricular cavity was "running off" through sinusoids into the arterial system and hence into the right atrium through the anomalous communication. Such findings suggest a certain possibility regarding the circulation in conditions wherein a coronary artery participates in an arteriovenous-like communication. The blood escaping abnormally through the anomalous coronary artery may be derived not only from that artery but also from other arteries with which it connects and even from the ventricular chambers.

Mention should be made of the developmental basis for the abnormal communication between the right coronary artery and the right atrium in the case reported herein. From the developmental analysis of the coronary vascular system made by Grant,[56] it is apparent that in the early stages of cardiac development the myocardium is nourished by blood which is brought to it through wide intertrabecular spaces. These exist in the myocardium and are continuous with the cardiac chambers. Later, the coronary veins and the coronary arteries develop and connect with the epicardial aspects of the intertrabecular spaces. Following this phase, the growth of the myocardium causes the intertrabecular spaces to be reduced in size to the caliber of capillaries. The intertrabecular spaces, however, maintain communication with the cardiac chambers and therefore represent, in the adult, the normal small communications

that exist between the coronary arteries and the cardiac chambers. In the case we have reported, the communication between the right coronary artery and the right atrium may therefore be considered a normal connection. The abnormality resides, therefore, not in the fact that connections of the coronary artery with the atrial cavity exist, but rather in the large size of the connections. Normally the communications should have been reduced to microscopic size. A similar view was expressed by Grant[57] for the abnormal communication between a coronary artery and a cardiac ventricle in the case previously reported by Wilson and Grant.[55]

SUMMARY

The case is reported of a woman having anomalous communication between the right coronary artery and the right atrium. Congestive cardiac failure was considered to be a complication of a left-to-right shunt. Dilatation of the anomalous arterial channel was complicated by the formation of a localized saccular aneurysm.

In creating a basis for an arteriovenous-like condition, the malformation in this case is representative of other cases in which the coronary arterial system communicates with the coronary venous system, a left or right cardiac chamber, or the pulmonary trunk. The continuous murmur often present in such conditions may be confused with patent ductus arteriosus or with other conditions which enter into the differential diagnosis of this malformation.

Surgical obliteration of arteriovenous-like fistulas in which the coronary arteries participate has been accomplished in a few reported cases.

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UNUSUAL ESOPHAGEAL ATRESIA WITH DISTANT MEMBRANOUS OBSTRUCTION OF THE ESOPHAGUS

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INCREASING experience with the surgical correction of esophageal atresia indicates that the majority of infants so afflicted may be saved if other severe anomalies are absent. Fortunately, most babies with esophageal atresia and associated tracheoesophageal fistula have no other important abnormalities. If such serious anomalies as congenital heart disease and malformation of the anus and rectum are present, they are readily recognized and do not often affect the initial treatment of the child.[5] Although rare, other anomalies do occur which affect the initial surgical management and the possibility of their presence should be constantly borne in mind. We have recently encountered such a combination and the unusual characteristics of the second abnormality prompted this report.

CASE REPORT

E. M., a 13-hour-old white female, was admitted to Jefferson Davis Hospital, Houston, Texas, on Dec. 31, 1955, because of difficulty in respiration, retraction of sternum, and transient cyanosis. These respiratory difficulties had ensued at birth in another hospital. Physical examination revealed a fairly well-developed, premature child, weighing 1,600 Gm., in mild respiratory distress. Increased respiratory effort with retraction of lower sternum was present, but cyanosis occurred only with crying. Expansion and breath sounds were equal bilaterally. Examination of the heart revealed marked cardiomegaly, a forceful precordial impulse with systolic thrill and a loud, harsh systolic murmur heard best in the left parasternal area and transmitted to the anterior and left posterior aspects of the chest. The liver was palpable below the costal margin. Initial laboratory studies were within normal limits. Roentgenograms of the chest revealed a markedly enlarged, globular-shaped heart. Electrocardiographic studies demonstrated right ventricular hypertrophy with severe ischemic changes. A diagnosis of congenital heart disease with an interatrial septal defect and/or interventricular septal defect was made. When further cyanosis and respiratory distress occurred, congestive failure was suspected and the child was digitalized. Some improvement followed but it soon became apparent that, in addition to respiratory distress, the child had excessive drooling of saliva and when oral feedings were begun regurgitation of all feedings occurred. Diagnosis of esophageal atresia was confirmed by the inability to pass a catheter through the esophagus and roentgenographic evidence of a blind upper esophageal pouch following instillation of 1 c.c. of Lipidol. No air was demonstrable in the gastrointestinal tract and it was felt that the atresia was not associated with tracheoesophageal fistula. Although some respiratory difficulty persisted, the general condition seemed improved enough by the third day of life to warrant surgical correction

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of the esophageal atresia. Accordingly, on Jan. 3, 1956, an extrapleural thoracotomy was performed. Exploration revealed a complete esophageal atresia of the usual type, with a blind upper segment just below the thoracic inlet and attachment of the distal segment to the posterior aspect of the trachea. The proximal aspect of the distal segment was narrow, firm, and cordlike at the point of tracheal attachment and the absence of the characteristic ballooning on inspiration of the distal segment and the absence of air in the gastrointestinal tract was felt to be on that basis. The attachment to the trachea was

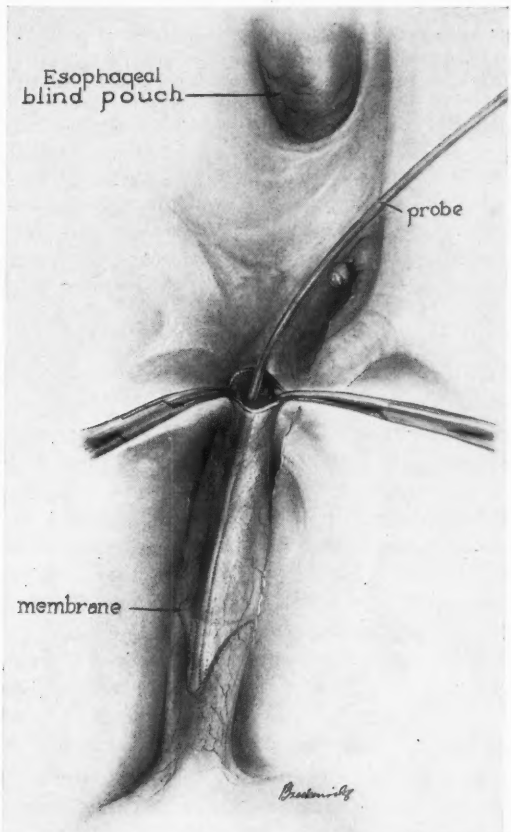

Fig. 1.—Artist's concept of the esophageal atresia. The tracheosophageal band has been divided and a proble in the distal segment demonstrates the pliable membranous obstruction.

divided and a small portion of the divided end sacrificed to secure an adequate lumen for anastomosis. A small probe was passed well into the distal limb and was thought to pass through the esophageal hiatus. Continuity of the esophagus was readily re-established without tension by an anastomosis employing two layers of fine silk sutures. A small, soft drain was placed retropleurally before closure of the chest wall. The child tolerated the procedure well and the immediate postoperative course remained good for approximately 48 hours at which time respiratory distress and cyanosis reappeared and the child suddenly died.

Post-mortem examination confirmed the suspected presence of severe congenital heart disease manifest by great dilatation and hypertrophy of the right atrium and ventricle, large atrial and ventricular septal defects, severe pulmonary stenosis, and a patent ductus arteriosus. The right pleural cavity was dry, the lung well expanded and the esophageal anastomosis intact. Upon removal of the esophagus and further inspection, the presence of a completely unsuspected congenital esophageal web was found in the lower esophageal segment approximately 2 cm. above the esophageal-gastric junction. Probing of the lumen of the distal segment, as was done at the time of surgical reconstruction of the proximal atresia, demonstrated that the pliable web could readily be stretched ahead of the probe as far as the stomach, thus giving an erroneous impression of patency of the distal segment (Fig. 1). The remainder of the necropsy findings were not remarkable with the exception of congestive changes in the lungs and a liver compatible with congestive heart failure.

DISCUSSION

Esophageal atresia may assume a variety of forms either with or without associated tracheoesophageal fistula; however, the majority occur in a characteristic pattern consisting of a blind upper segment and a lower pouch connecting with the trachea. Because of the frequency of this type of anomaly, treatment has become relatively standardized with significant improvement in results.

Despite the large number of cases of tracheoesophageal fistula now reported in the literature, the occurrence of a membranous obstruction of the esophagus in association with complete atresia and tracheoesophageal fistula has not been described. Theoretically, a membranous form of atresia might occur in the esophagus as it does in other portions of the gastrointestinal tract but membranous obstruction per se is quite rare. MacKenzie,[7] Tenon,[12] Rossi,[8] and Abel[1] have described membranous obstruction and it is likely that many of the cases of congenital stenosis represent incomplete webs or membranes. The only recorded instance of membranous obstruction associated with tracheoesophageal fistula was the interesting case of Schwartz and Dale.[9] In this case, continuity of the esophagus was not interrupted and the fistulous connection was distal to the web. Gross[3] encountered a stenotic area in the distal segment of one case in their large series of atresia and associated tracheoesophageal fistula, but no case of membranous obstruction associated with proximal atresia and tracheoesophageal fistula has been reported.

The presence of a weblike membrane in the distal segment of our case was completely undetected by the usual maneuver of passing a probe or catheter into the stomach. The elasticity of the structure allowed it to be readily pushed ahead of the probe, a maneuver we were able to duplicate with the post-mortem specimen (Fig. 1). Fortunately, the outcome of this particular case was not influenced by this additional anomaly but, had the child survived, an additional operative procedure would have been required. The existence of a second obstruction was considered and sought because of the absence of air in the gastrointestinal tract but the demonstration of the cordlike nonpatent attachment to the trachea, a condition previously described,[4] was considered an adequate explanation for the absence of air. The secondary obstruction could have been readily corrected at the time of the original procedure by

everting the distal segment or by a small esophagotomy at a lower level with resection of the web. The demonstration of aspirated bile through a catheter into the stomach would have circumvented the error and we have since added this maneuver to the management of these cases. Aspiration of the distended stomach may also be carried out at this time. Since hypertrophic pyloric stenosis and duodenal obstruction are frequently associated anomalies,[2, 6, 9, 10, 11] demonstration of bile in the catheter is evidence of patency of the pylorus and upper duodenum.

SUMMARY

An unusual case of esophageal atresia with bandlike attachment to the trachea associated with complete membranous obstruction in the distal esophageal segment is presented. At operation for tracheoesophageal fistula a catheter should be introduced into the distal esophageal segment and into the stomach to determine the presence of such a membranous obstruction, which can be easily corrected if found. The aspiration of bile will also exclude hypertrophic pyloric stenosis and duodenal obstruction, which are occasional associated anomalies.

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SECONDARY NEOPLASMS OF THE MEDIASTINUM

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DEVELOPMENTS and progress in the field of oncology have frequently resulted from investigations and considerations of the natural history of neoplastic diseases. Since some secondary neoplasms of the mediastinum are a part of the clinical course of several surgically treated malignant tumors, and since they have not been studied extensively, an investigation of them was initiated in 1951.

MATERIAL AND RESULTS

Protocols of autopsies performed in general hospitals over a period of years offered the best source of material for such a study. From August, 1895, to Dec. 31, 1950, 9,248 autopsies were performed by physicians in the Pathology Department of the Presbyterian Hospital. The anatomic diagnoses of each autopsy were examined in order to learn the presence of a neoplasm. In such cases, the gross description and report of microscopic examinations were scrutinized to learn of possible mediastinal involvement. Lesions of the lymph nodes, such as Hodgkin's disease and lymphosarcoma, were not included in the study. In these cases of generalized disease, it is difficult to decide at autopsy which is the primary or secondary lesion.

There were 337 protocols which recorded neoplasms that had metastases involving the mediastinum. In Table I, the primary neoplasms from which these metastases originated are listed. Carcinoma of the breast, bronchus and lung, esophagus, and stomach, the most common malignant diseases of both sexes, are at the top of the list. The secondary neoplasms of the bronchus and lung were placed in one group because their exact origin cannot always be determined. Forty-five types of primary tumors are included.

As a sidelight, a tabulation of the cases of secondary neoplasms, by decades in the period studied, was made. Table II demonstrates an increasing number of the primary neoplasms with such metastases in the breast, bronchus, and lung. There was an average of 50 more autopsies a year in the 1940's than in the 1930's. This would account for only a portion of the increased incidence of cases of metastases in the last decade studied.

The nomenclature of Rouviere[1] was used to describe the location of the metastases as recorded in the protocols. The lymph nodes, as well as other structures in the mediastinum most frequently involved by the secondary neoplasms,

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TABLE I. INCIDENCE OF SECONDARY MEDIASTINAL NEOPLASMS

PRIMARY NEOPLASM	NUMBER OF CASES OF SECONDARY NEOPLASMS IN THE MEDIASTINUM
Carcinoma of breast	73
Carcinoma of bronchus and lung	60
Carcinoma of esophagus	35
Carcinoma of stomach	35
Carcinoma of pancreas and colon, each	12
Carcinoma of urinary bladder	10
Carcinoma of prostate	9
Carcinoma of gall bladder, kidney, and unknown etiology, each	7
Carcinoma of uterus and ovary, each	6
Carcinoma of liver and thyroid, each	5
Malignant melanoma	5
Sarcoma of extremities, etc.	5
Sarcoma of retroperitoneal region	4
Carcinoma of rectum	3
Carcinoma of extrahepatic bile ducts	2
Carcinoma of skin and tongue, each	2
Osteogenic sarcoma of femur	2
Sarcoma of unknown origin	2
Carcinoma of trachea, pharynx, larynx, buccal mucosa, left ureter, penis, vagina, each	1
Sarcoma and leiomyosarcoma of esophagus, each	1
Reticulum-cell sarcoma of stomach	1
Melanosarcoma of liver	1
Mixed tumor of kidney, left	1
Myeloma	1
Fibrosarcoma of unknown etiology	1
Neuroblastoma of spinal nerve roots	1
Adenocarcinoma and sympathogonioma of adrenal gland, each	1
Testicular tumors: seminoma type (?), malignant, teratoma, and carcinoma, each	1
Total	337

TABLE II. INCIDENCE OF SECONDARY MEDIASTINAL NEOPLASMS BY DECADES

	DECADES					
	1895-1910	1911-20	1921-30	1931-40	1941-50	TOTALS
Number of Autopsies	*2,133*	*1,336*	*1,856*	*1,722*	*2,201*	*9,248*
Breast	0	4	11	15	43	73
Bronchus and lung	3	0	10	13	34	60
Esophagus	8	1	14	7	7	37
Stomach	4	1	7	12	12	36
All Others	14	10	20	34	53	131
Totals	29	16	62	81	149	337

are given in Table III. Only the cases of metastases of the more common primary lesions are included. In some cases it was impossible to decide which lymph node group were involved as the protocols referred only to mediastinal lymph nodes and not to their exact location. For the sake of completeness, these cases, worthy of minor consideration, are enumerated at the end.

In addition to learning which of the mediastinal lymph nodes contained metastases, an attempt was made to determine in which cases the involvement could have been recognized clinically, as by gross examination at the time of a thoracotomy. Several criteria were used for making these decisions. One was the consistency and, another, the size of the lymph nodes involved. The

TABLE III. LYMPH NODES AND STRUCTURES INVOLVED BY SECONDARY NEOPLASMS IN THE MEDIASTINUM

	PRIMARY NEOPLASM			
	STOMACH	ESOPHAGUS	LUNG AND BRONCHUS	BREAST
Number of cases	*36*	*37*	*60*	*73*
Juxta-esophageal LN	6	16	7	13
Thoracic duct	2	3	2	2
Juxta-aortic LN	11	4	12	16
Pulmonary root LN	1	9	17	21
Anterior mediastinal LN	3	1	14	18
Pericardium	0	–	14	19
Right laterotracheal LN	23	14	38	40
Inter-tracheobronchial LN	19	11	32	32
Left laterotracheal LN	21	12	33	41
Mediastinal LN	10	5	24	31

LN—lymph node.

latter is not necessarily a satisfactory criterion because it is well known that lymph nodes are enlarged by infections. However, used in conjunction with the consistency of the node, as by palpation, it is valuable. In Table IV, these data are summarized.

TABLE IV. CLINICAL DATA CONCERNING LYMPH NODES

	NO. OF CASES	GROSSLY INVOLVED	GROSSLY INVOLVED (?)	SEEN ON X-RAY
Stomach	36	31	0	0
Esophagus	37	32	5	1
Breast	73	60	1	8
Bronchus and lung	60	56	0	16

Another attempt was made, during the analysis of the data, to decide which of the secondary neoplasms could be seen on a routine thoracic roentgenogram. Two criteria were utilized for a positive conclusion. One was the size of the involved node, or nodes, and the other was its location. According to McCort and Robbins,[2] the tracheobronchial and right anterior mediastinal chains are more easily seen than are most of the other nodes of the mediastinum on such a roentgenogram. When the involved node, or nodes, was in the above groups and 5 cm. in diameter, or larger, it was concluded that it could be seen. According to this evaluation, approximately 25 per cent of the secondary neoplasms of bronchiogenic carcinoma could be seen. However, in only one case of carcinoma of the stomach and esophagus could an enlarged node be seen. If these criteria are accurate, less than 15 per cent of the mediastinal metastases from carcinoma of the breast could be outlined on a thoracic roentgenogram.

DISCUSSION

Only recently has there been an attempt by clinicians to utilize the anatomist's descriptions of the lymphatic system of the mediastinum.[10 11 12] The results of the present investigation are published to focus more attention not only on the mediastinum but its chains of lymph nodes and their position in the spread of malignant disease.

In Table I, a large number of primary neoplasms are found to metastasize to the mediastinum. Previous lists[3, 7, 8, 13] have been more limited in number. A number of primary neoplasms of the abdomen and pelvis, as well as of the head and extremities, are present in this series. As a result of this investigation, it is apparent that practically any carcinoma or sarcoma in or on the human body is capable of spreading to the mediastinum. In this study, as in the series of Abrams and his associates[9], carcinoma of the breast was the first in order of incidence and carcinoma of the lung was second.

The increasing incidence of primary carcinoma of the lung in Table II is obvious and confirms the opinions of these interested in this disease in this coun try and abroad.

Table II also authenticates the opinion that neoplastic disease has been increasing. Undoubtedly the significant factor is that more people are living to an older age.

Probably the most significant data are compiled in Table III. The marked incidence of involvement of the tracheobronchial lymph nodes is noteworthy. The nodes of the tracheal bifurcation, or inter-tracheobronchial lymph nodes, are a crossroad for lymphatic or lymphoid relays. The large number of these nodes involved by secondary neoplasms of the stomach is an exception to previous pathologic studies and reports.[4, 13]

It is remarkable that a large number of the involved lymph nodes could be distinguished by their gross appearance (Table IV). However, only a few cases had involvement which could be diagnosed on the usual posteroanterior film. The size of 5 cm. (in diameter), for the lymph nodes or lymph node masses, was used because a lesion of this size would probably be distinguished from the density due to the spine, cardiac shadow and other mediastinal structures. The routine of McCort and Robbins[2], and other radiologists, of utilizing a barium swallow and a Bucky film, or planigrams, should increase the number of clinically diagnosed metastatic lesions of the mediastinum.

SUMMARY AND CONCLUSIONS

This is the first known study of the incidence of secondary neoplasms of the mediastinum and of the lymph nodes involved by them.

Clinicians, pathologists, and radiologists should be more exacting in their terminology in describing the lymph nodes of the mediastinum. Designating these nodes as mediastinal is not satisfactory for the future consideration and management of patients with such metastases.

Any malignant neoplasm may metastasize to the lymph nodes of the mediastinum. The most frequently found metastases are from carcinomas of the breast, lung, esophagus, and stomach.

The fact that many of the involved nodes of the mediastinum can be distinguished grossly, as at the time of thoracotomy, has clinical value. However, microscopic metastases are frequently present in grossly uninvolved nodes.

Unfortunately, the present routine used in many hospitals of taking a posteroanterior chest film to decide if there are intrathoracic metastases is insufficient. This study demonstrates that in relatively few instances, metastases of the mediastinum can be seen with such a method.

One of the avenues for further progress in oncology lies in more detailed studies and in better management of secondary neoplasms of the mediastinum.

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RADIATION THERAPY FOR RECURRENT CARCINOMA IN THE BRONCHIAL STUMP

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THE surgical approach to bronchogenic carcinoma has left in its wake an increasing number of recurrences in the bronchial stump. This report details a course of management, combining surgery and three full courses of radiation therapy, which allowed such a patient to lead a productive life for 4½ years following the definitive diagnosis of his malignancy.

CASE REPORT

The patient (G. C. M.), a 57-year-old white man, was first seen in April, 1952, with a history of chronic respiratory infection dating back to 1946. He had been a heavy cigarette smoker for 25 years. His cough had increased progressively over the previous 6 weeks. He had had fever, dyspnea, and occasional hemoptysis. Râles and decreased breath sounds were detected at the right base. Roentgenograms and fluoroscopy of the chest demonstrated an enlargement of the right root and partial atelectasis of the right lower lobe (Fig. 1). Bronchoscopy revealed a large mass in the right main bronchus just below the carina. Pathologic examination of biopsy material removed at bronchoscopy showed epidermoid carcinoma.

On April 25, 1952, a right pneumonectomy was performed during which the surgeon noted tumor tissue at the line of bronchial transection at the carina (Fig. 2). The hilar and mediastinal nodes were enlarged; however, the pathologist reported no tumor found in the nodes removed proximal to the transection. Postoperatively the patient gained weight, had less dyspnea, and returned to his work as an executive.

In May, 1953, weight loss, cough, and hemoptysis recurred. Bronchoscopy on Aug. 7, 1953, 16 months post-pneumonectomy, revealed recurrent tumor in the right main bronchial stump (Fig. 3). Tumor partially occluded the left main bronchus as well. In an effort to clear the airway, the bronchoscopist removed as much tumor as possible.

On Aug. 14, 1953, the patient was started on a course of radiation therapy with the carina as the center of rotation and utilizing a 10 × 10 cm. port with a focal axis distance of 100 cm. One million volt radiation was employed at 3 Ma. and HVL of 9.0 mm. of copper. Over a period of 26 treatments and 38 days, a tumor dose of 5,200 r was delivered. The patient tolerated the treatment well, showing progressive improvement and experiencing only minimal reddening of the skin over the sternum (Fig. 4).

The patient did very well thereafter, complaining only of slight dyspnea but no cough, fever, or hemoptysis until February, 1955, when he again raised blood-streaked mucus. On Feb. 18, 1955, bronchoscopy again revealed recurrent tumor in the bronchial stump on the right, partially occluding the left main bronchus (Fig. 5). The bronchoscopist removed as much tumor as feasible and the patient was begun on a second course of radiation

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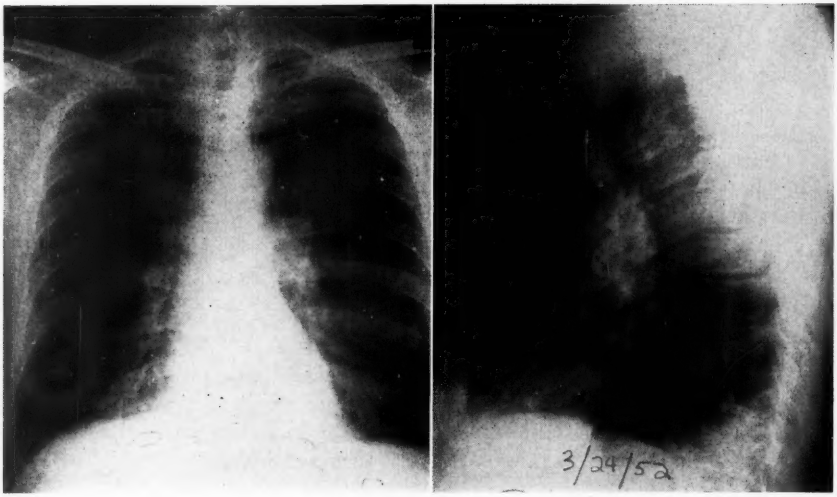

A. *B.*

Fig. 1.—PA (*A*) and lateral (*B*) chest films made on March 24, 1952, demonstrate atelectatic segment at right base posteriorly. Fluoroscopist noted shift of heart to right on inspiration.

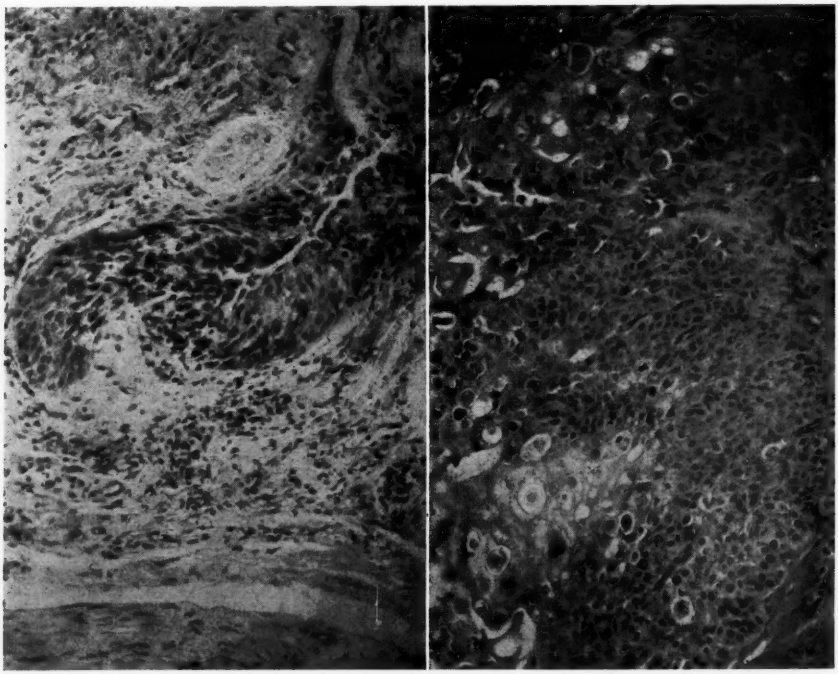

Fig. 2. **Fig. 3.**

Fig. 2.—Section from surgical specimen shows only demonstrable focus of microscopic invasion. Remainder of tumor was papillary in nature.

Fig. 3.—Biopsy of recurrence on Aug. 7, 1953, shows a fibrous stroma infiltrated with epidermoid cells. The cells appear fairly mature but show some mitoses and variations.

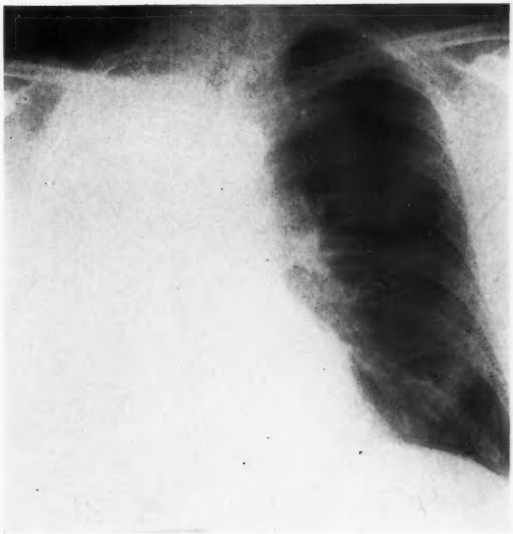

Fig. 4.—PA roentgenogram of chest made on Sept. 15, 1953, 17 months after pneumonectomy and on completion of first course of radiation shows a normal post-pneumectomy chest.

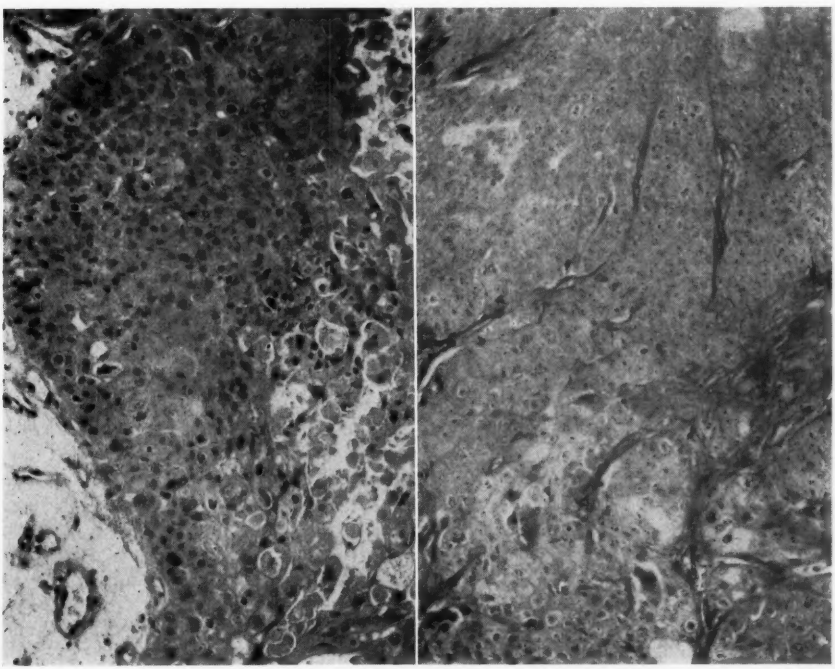

Fig. 5. Fig. 6.

Fig. 5.—Biopsy of second recurrence on Feb. 18, 1955, again shows fairly mature invasive epidermoid carcinoma.

Fig. 6.—Biopsy on June 29, 1956, demonstrates similar recurrent tumor.

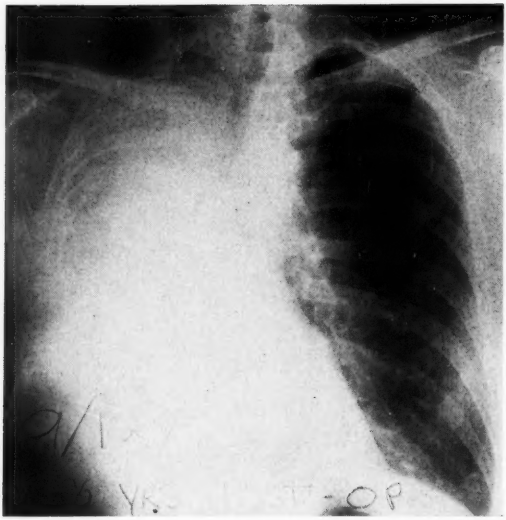

Fig. 7.—PA roentgenogram of chest made on Sept. 12, 1956, 4½ years after operation and after administration of 15,200 r to the carina, shows only normal post-pneumonectomy changes.

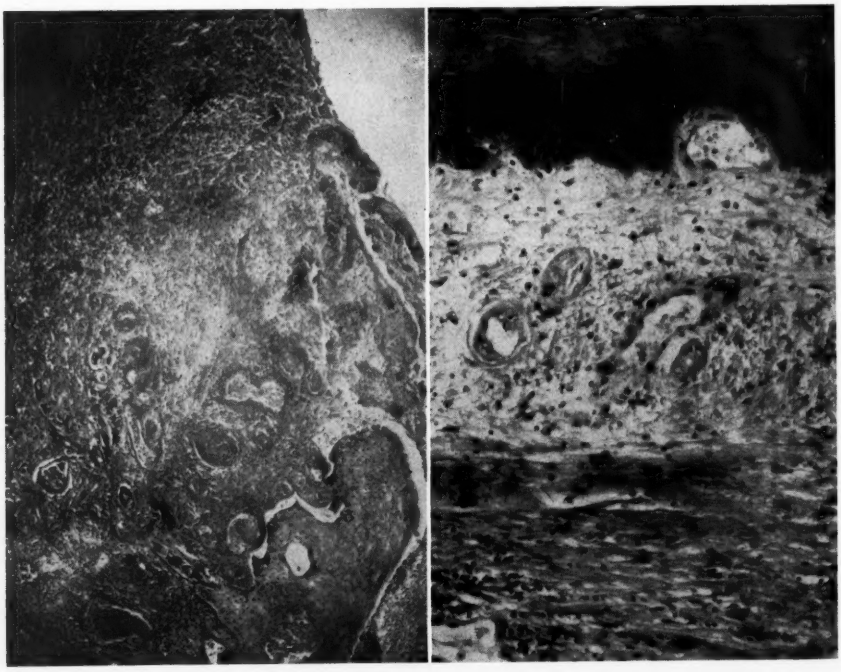

Fig. 8. Fig. 9.

Fig. 8.—(A 16631) Section through margin of carinal ulcer shows hyperplastic squamous epithelium, vascular changes, necrosis, and acute inflammation. No tumor cells can be identified.

Fig. 9.—(A 16631) Extensive squamous metaplasia of tracheal mucosa with some chronic inflammation and vascular changes presumably the result of radiation.

therapy on Feb. 23, 1955. The same factors as described above were utilized and over a period of 26 treatments in 36 days, 5,000 r was delivered to the carina. The patient noted diminution of cough and dyspnea and was able to continue at work throughout therapy.

In June, 1956, cough returned along with hemoptysis and dyspnea. Once again the bronchoscopist cleaned out as much tumor as technically possible and obtained pathologic proof of recurrent epidermoid carcinoma (Fig. 6).

The hazards of further radiotherapy were weighed against the patient's otherwise hopeless outlook and on July 10, 1956, a third course of radiation therapy was started, using the factors described above. Over a period of 27 treatments and 37 days, 5,000 r was again delivered to the tumor. The patient continued at his job with some exertional dyspnea and cough, although less than prior to his last course of treatment (Fig. 7).

On Oct. 21, 1956, 4 years and 6 months following the original surgery, an upper respiratory tract infection developed and after an initial response to antibiotics and bed rest, he died on Oct. 22, 1956, with a massive pulmonary hemorrhage.

At post-mortem, an area of necrosis, measuring 4 cm. by 5 cm., was found in the tracheobronchial tree at the location where recurrent tumor had been seen at bronchoscopy prior to therapy. This was thought to represent the source of terminal hemorrhage. No definite evidence of residual tumor cells was found on detailed pathologic investigation of sections through the area of necrosis or elsewhere (Figs. 8 and 9). A moderate degree of radiation fibrosis was present in the remaining lung as well as some pleural fibrosis. Marked tracheitis was also seen.

DISCUSSION

Despite vigorous efforts to improve and refine methods of diagnosis and treatment, bronchogenic carcinoma continues to take a toll of approximately 28,000 lives annually and is the second most frequent cause of death due to neoplasm among males in the United States today. In wholly unselected series, the 5-year survival of patients with an established diagnosis of bronchogenic carcinoma is estimated at less than 5 per cent.[1]

Radiation therapy following surgery is seldom prescribed for patients suffering with bronchogenic carcinoma with the expectation of achieving cure, but rather as a palliative measure. Usually, the intent of the radiotherapist is to relieve bronchial obstruction and secondary peripheral infection, superior mediastinal obstruction, pain, cough, or acute pulmonary osteoarthropathy.[2] There is considerable debate as to whether radiation therapy actually does increase longevity even when palliation is achieved.[3]

Habein and associates[4] of the Mayo Clinic recently reported on 18 patients with recurrence in the bronchial stump, several of whom received radiation therapy. We have had 4 recent cases of such recurrence, one of which is reported. The marked palliation obtained in this case, employing surgery and repeated courses of radiation therapy, has encouraged us to attempt a similar plan in other cases. It is too early to evaluate our results in the 3 cases not reported here.

The ease with which this patient tolerated his re-treatment with radiation has raised the question as to whether re-treatment might not also be of value in situations other than bronchial stump recurrence. A dilemma occasionally arises in the patient who has had 18 to 24 months' arrest of bronchogenic carcinoma following radiation therapy and who has tolerated his original course

of treatment well. Lacking evidence of recurrence, should such a patient be re-treated on the basis of the established experience that tumor cells almost assuredly persist?

SUMMARY

We have presented a patient who received 15,200 r to the carina over a 3-year period. His death was due to hemorrhage, secondary to slough of necrotic tissue in which no tumor was found. Our experience in this case has encouraged us to believe that radiation therapy is of value in the management of postoperative bronchial stump recurrence. In addition, our experience has shown that, in selected cases, a second course of radiation therapy can be tolerated and should be investigated further in situations other than bronchial stump recurrence as to its potential use in the long-term control of bronchogenic carcinoma.

We would like to thank Dr. Roger Terry, Associate Professor of Pathology, and Dr. Marion Emerson, Instructor in Medicine, both of the University of Rochester School of Medicine and Dentistry, for their guidance and assistance in preparing this paper.

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PERICARDIAL FAT NECROSIS: AN UNUSUAL ENTITY

A Case Report

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A REPORT by Jackson, Clagett, and McDonald of the Mayo Foundation[1] describes 3 cases of a new disease entity, which these authors termed pericardial fat necrosis. These authors stated that a review of the literature revealed no other such cases reported. In view of the paucity of literature on this entity, the following case appears to be worthy of reporting. Furthermore, the previous essayists on this disease entity made a postulation that obesity, which was present in all 3 of their cases, was probably a prerequisite of this disease. Our case would indicate that this is not a constant prerequisite. Furthermore, the cases reported from the Mayo Foundation were in the fifth, sixth, and seventh decades of life. The case reported herein occurred in a patient in the third decade of life.

CASE REPORT

A 23-year-old white man was admitted to a nearby hospital on February 23, 1957, with the chief complaint of pain in the left chest. The onset occurred 24 hours prior to admission at which time he was seen by his family physician. He described the pain as severe precordial pain radiating into the neck and left shoulder. He was pale and appeared clinically to be in mild shock. It was the opinion of the physician at that time that, despite his age, a coronary occlusion was probable. The patient was slightly dyspneic, but not cyanotic, when admitted to the hospital. Physical examination at the time of this hospital admission was not remarkable. He described pain over the left anterior chest wall and slightly less pain and tenderness over the left upper quadrant of the abdomen. The blood pressure was 124/78 mm. Hg. The pulse was slightly rapid, but the rhythm was regular. There were no abnormal heart sounds. An electrocardiogram was taken on the day of admission and revealed a normal record.

The laboratory findings upon admission revealed a normal urinalysis. The hemoglobin and red blood count were within normal limits. The white blood count was 12,250 with 54 per cent segmented cells, 17 band cells, 24 lymphocytes, 4 monocytes, and 1 eosinophil. The pain gradually increased. Twenty-four hours after admission he was seen by a medical consultant. The physical findings were essentially the same as on admission. It was the opinion of the medical consultant at that time that the patient probably had epidemic pleurodynia. A roentgenogram of the chest was ordered. Because of the patient's acutely ill condition, this was a bedside roentgenogram. On this examination a density was noted in the left lower lung. It was noted by the radiologist that the shadow obscured visualization of the left heart border and the left diaphragm. A follow-up roentgenogram was advised and this was taken 2 days later (Figs. 1 and 2). On this film the definition was much better than on the first study. A round, discrete shadow was described in the left anterior base. Fluoroscopy was carried out at this examination and the radiologist noted that the mass could not be separated from the cardiac silhouette. During the next few days, on antibiotic therapy and analgesics, the patient's symptoms improved and he was discharged on March 2, 1957.

Because of persistence of the anterior chest pain he was admitted on March 19, 1957, to Aultman Hospital. On admission he was afebrile. The white blood count was 7,300 cells

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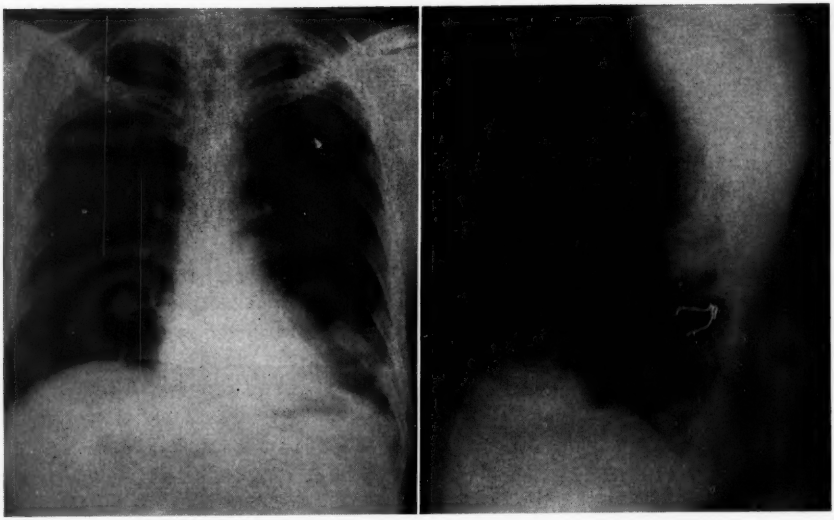

Fig. 1. Fig. 2.

Figs. 1 and 2.—Posteroanterior and left lateral roentgenograms of the chest demonstrating density near the apex of the heart due to pericardial fat necrosis.

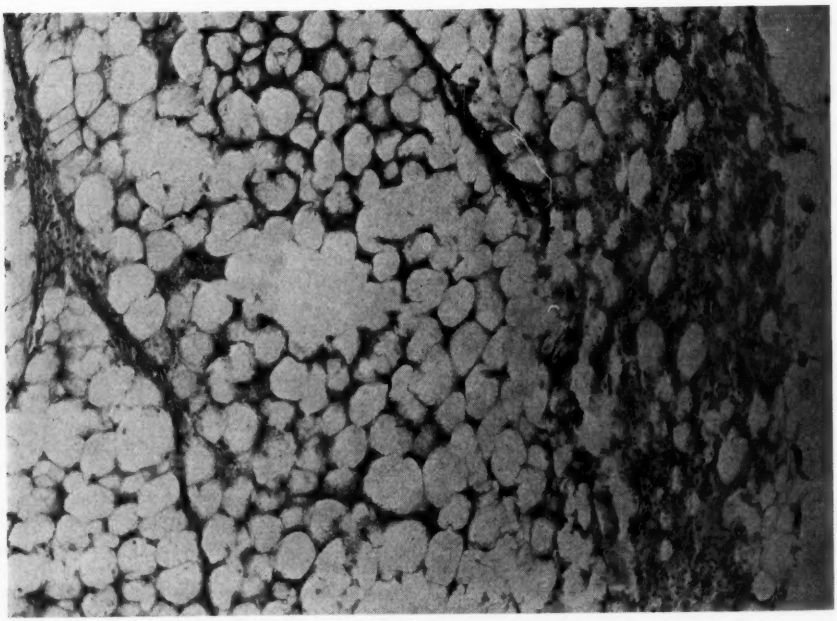

Fig. 3.—Necrosis of fat with formation of pseudocysts. Xanthoma cells are present at the periphery. (Hematoxylin and eosin; ×109.)

with a normal differential. A roentgenogram of the chest was taken just prior to surgical exploration. This demonstrated essentially the same findings as on the roentgenogram taken at the other hospital soon after his admission there. On March 20, 1957, an exploratory thoracotomy was performed through a classical left posterolateral approach. An indurated mass, measuring 7.0 by 4.5 by 2.0 cm., was found attached to the pericardium near the apex of the heart. It was adherent to the anterior chest wall and the lingula of the lung by fibrinous adhesions. The areas of adhesions showed acute inflammatory changes.

The mass itself was attached to the pericardial surface by a broad flat surface, measuring 7.0 by 4.5 cm. It was coarsely lobulated and was light brown in color. Grossly, it resembled an adrenal gland. This mass was dissected from the anterior chest wall, lingula, and the pericardium. The bleeding points were ligated and the chest was closed with drainage in the routine manner. The patient was discharged on March 27, 1957, on the ninth postoperative day. His postoperative course was uneventful and he was very promptly relieved of his precordial chest pain. It should be noted here that the patient was 5 feet 10½ inches tall, and weighed 155 pounds.

The microscopic examination of the specimen is as follows*: The specimen is composed of fat cells, many of which show granular degeneration or necrosis. In places the cell membranes are indistinct or are ruptured so that small pseudocysts are formed. No granulomatous reaction is present. The fat is divided into lobules by delicate strands of connective tissue which show lymphocytic infiltration. At the periphery of the specimen there are numerous groups of xanthoma cells, associated with mild fibroblastic proliferation and exudation of lymphocytes (Fig. 3).

COMMENT

We believe this is the same clinicopathologic entity described by Jackson, Clagett, and McDonald. However, we would point out that our patient was several years younger than any of the 3 patients reported by these authors, and, in contradistinction to the obese patients in which they found this disease, our patient was a rather thin person. The previous authors attempted to relate the etiology of this condition to a similar condition found in the appendices epiploicae and also in the omentum. Our case has not elucidated the pathogenesis of this unusual and apparently rare condition. In view of the cases thus far reported, it would appear that surgical excision of the necrotic, inflamed mass gives prompt relief. However, this may not prove to be the ideal therapeutic approach if more is learned about this disease.

SUMMARY

1. A case of fat necrosis involving the parietal pericardial fat is reported.

2. To the best of the author's knowledge, this is only the fourth such case to be reported in the literature.

3. The outstanding clinical features of this condition are acute anterior chest pain, resembling the pain of coronary occlusion. On roentgenograms there is an area of increased density adjacent to the left border of the pericardium.

4. Histologically, the lesion resembles fat necrosis found in other situations.

5. The etiology of this condition remains obscure.

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*The microscopic slides were reviewed and described by Dr. Simon Koletsky, Professor of Pathology, Western Reserve Medical School, Cleveland, Ohio.

INTERNAL MAMMARY VENOGRAPHY

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THE value of intra-osseous venography in the demonstration of certain venous pathways, which hitherto partly or completely escaped roentgenographic visualization by conventional means, is now firmly established and confirmed by numerous reports. In the following report some aspects of this method will be discussed.

TECHNIQUE

The usual tests concerning possible idiosyncrasies to either the contrast substance or the local anesthetic agent must be taken. Full cooperation by the patient is ensured by explaining the procedure and mentioning the moderate amount of local discomfort experienced during the injection. Adequate sedation is obtained with meperidine hydrochloride (75 to 100 mg.) and secobarbital sodium (0.1 gram). Both drugs are administered one hour prior to the procedure.

The patient is supine on the x-ray table with both arms elevated and the correct positioning is verified with a scout film. A cutaneous wheal is raised with 1 per cent procaine hydrochloride in the midline of the sternum at the level of the fourth rib anteriorly. Additional infiltration of the sternal periosteum is advisable. A 16-gauge Rosenthal bone marrow aspiration needle is then introduced with a rotary motion into the medullary cavity of the sternum. The needle should be inclined approximately 30 degrees so as to direct its tip cephalad. The aspiration of a small amount of bone marrow as well as the accompanying sensation of local "pressure" speak in favor of adequate placement of the needle.

The radiopaque substance used in the present study was a 50 per cent solution of Hypaque† (3,5 diacetamido-2,4,6 triiodobenzoate) which, in contradistinction to other previously employed media, was found to exhibit a remarkably low incidence of ill effects, to be much less painful to the patient, and to yield roentgenograms of better contrast. An ordinary 10 c.c. manual syringe is connected to the needle by means of a piece of plastic tubing. The injection of contrast material (8 to 10 c.c.) is carried out under constant and moderate pressure. The patient is invited to breathe regularly through the open mouth

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†Hypaque was made available through the Research Department of Winthrop-Stearns, Inc.

since forceful respiration and periods of apnea (Valsalva exercise) may lead to poor and thus unsatisfactory filling of the veins. One roentgenogram is usually all that is required as long as the film is exposed *during* the injection of the last 1 to 2 c.c. of contrast medium. Delayed exposures do indeed reveal only a minute residual deposit of contrast substance within the medullary cavity of the sternum. If the films are found to yield satisfactory information, the needle may be withdrawn and the patient sent back to the floor. No special aftercare is required. Internal mammary venography was in several instances performed on ambulatory patients who, after completion of the procedure, were discharged home without incidence.

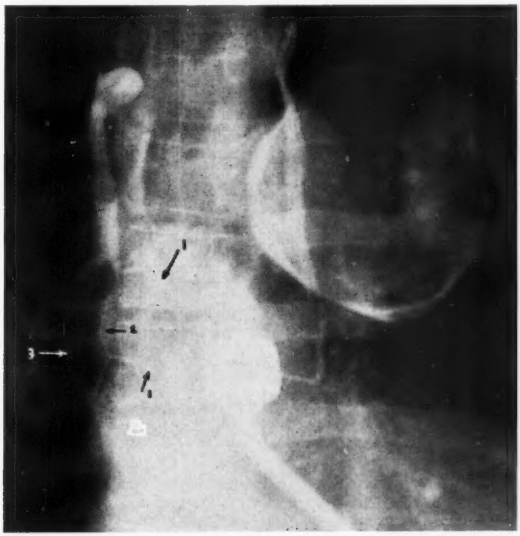

Fig. 1.—*Normal* internal mammary vein pattern. Case of cystic teratoma of the anterior mediastinum. *1,* Small veins draining the medullary cavity of the sternum and continuing in longitudinal direction, *2. 3,* Less densely filled portion of internal mammary vein.

DISCUSSION

Internal mammary venography was carried out in a series of 50 unselected patients with primary breast disorders of benign and malignant nature. Included also were patients with postoperative metastatic carcinoma to axillary and supraclavicular nodes, and with or without evidence of involvement of the chest wall.

Although the limited number of patients examined by this method does not allow statistical conclusions, there are nevertheless several observations worthy of comment.

The injected contrast medium readily escapes, under normal conditions, the medullary cavity through several small veins which join a rather large venous channel on each side of the sternum (Figs. 1 and 2). These veins follow a

course parallel to the sternal edge and their anatomic location corresponds to the one occupied by the internal mammary veins. The caliber of these veins remains more or less equal and filling in proximal direction can usually be followed up to their point of entry into the distal portion of the innominate veins, while the visualization in distal direction may be variable.

The small branches which are seen to drain the medullary cavity of the sternum may not always immediately join the internal mammary veins but are occasionally seen to run in a longitudinal direction along the sternal border.

Fig. 2.—*Abnormal* internal mammary vein pattern. *A*, Area of obstruction caused by enlarged metastatic lymph nodes in the right second intercostal space. Deviation of blood flow over collaterals to ipsilateral, *B* and opposite vein, *C*, *D*. Left internal mammary vein essentially normal.

It is important to recognize this fact and not to mistake these veins for narrowed segments of the internal mammary veins, especially if less dense filling of these latter structures has been obtained (Fig. 1). There may, on the other hand, exist a real narrowing of the internal mammary vein itself, independent of any extrinsic pressure, which, as observed in several of our cases, usually occurs in the region of the third costal cartilage and mostly on the right. That such narrowing does not merely represent a roentgenographic artifact, due to insufficient filling of the vein with contrast medium, has repeatedly been confirmed by surgical exploration of the respective area.

Since the internal mammary vessels are normally maintained in a more or less fixed position, frank displacement of these veins by retro- or parasternal masses will rarely occur. There will rather be an encroachment upon the wall of these vessels with resulting narrowing of their lumen. In more advanced cases (Fig. 2), there may be complete block of the flow of blood within the internal mammary vein at the level of the lesion with accompanying development of collateral venous channels. Some degree of marrow stasis due to delayed outflow from the sternal marrow cavity may also be observed and is a prominent feature in cases of bilateral internal mammary vein obstruction. The collateral channels may communicate again with the involved vein at a level above the obstruction or with intercostal veins and even with the internal mammary vein on the opposite side. Furthermore, there is increased filling with contrast medium of the distal portion of the internal mammary veins.

The histologic evaluation of material removed by open biopsy from the site of demonstrable or suspected obstruction leads to the conclusion that various disorders may partially or completely block the internal mammary vein at any given level. Such lesions consisted of prominent parasternal fat pads, fibrous plaques following external irradiation, abnormally narrow vein segments, and metastatic cancer involving the internal mammary nodes.

In other words, internal mammary vein obstruction per se does not speak in favor of carcinomatous involvement of internal mammary lymph nodes even if the presence of cancer, mostly of the breast, has definitely been established. Conversely, the demonstration of a partial or complete block of the internal mammary vein in patients who are clinically "cured" from their malignant process may indicate the existence of a hidden and unrecognized focus of metastatic carcinoma. What, furthermore, can be learned from these observations is that internal mammary venography enables the surgeon to biopsy the correct site of possible metastatic involvement, be it prior to surgery or after treatment. Hence, not only a better selection but also a more adequate follow-up of certain patients should be possible with the initiation of proper secondary therapy at an earlier and thus more favorable time.

It is doubtful whether internal mammary venography would be of any benefit in the study of certain anterior mediastinal tumors when their location, relationship to the surrounding structures, and other characteristics are concerned. It can be anticipated that most lesions in that particular location will not encroach upon the internal mammary veins (Fig. 1).

SUMMARY

The technique of internal mammary venography is described and the advantages as well as limitations of this procedure are emphasized.

The more frequent use of this method in the follow-up study of clinically "cured" patients may well lead to a better evaluation of the therapeutic results in breast cancer.

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A COMPACT SUCTION APPARATUS FOR CHEST DRAINAGE

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THE 3-bottle suction apparatus for chest drainage catheters which is described in textbooks of thoracic surgery[1, 2, 3] is a simple, effective apparatus, but it has a number of defects. The three bottles, plus a suction pump, occupy a considerable amount of space and they are difficult to move. Nurses find it troublesome to empty the drainage bottle but measurement of the fluid drainage will not be accurate unless the bottle is emptied. The connecting rubber tubes must be watched carefully to avoid kinking and obstruction. The bottles are easily tipped over and broken, thus connecting the pleural space with atmospheric air. To correct some of these defects and to make the whole apparatus more convenient for the nursing staff, we have modified the 3-bottle suction system into a compact, rugged unit of simple design.†

The three bottles of the unit—the drainage bottle, the air leak indicator, and the vacuum regulator bottle—are all small and arranged compactly on a substantial metal platform which has room for one Stedman pump (Figs. 1 and 2). The entire apparatus stands conveniently under the hospital bed. The Stedman pump maintains a negative pressure, the degree of which is regulated by the depth of the vent tube in the column of mineral oil. For a small volumn tube, such as this one, mineral oil has been better than water because it does not evaporate. If there is an air leak from the lung, this is indicated by bubbling from the small tube of the air leak indicator and this indicator bottle also serves as a water seal should suction fail. Adjustments in the amount of negative pressure can be made easily by changing either the level of the oil or the depth of the tube beneath the oil. The use of a larger inside tube in the air leak indicator prevents water from being sucked over into the drainage bottle when the patient makes a strong inspiratory effort.

All parts of the apparatus are easily disassembled for cleaning or replacement. The metal parts are stainless steel. The apparatus is designed so that only short, relatively straight, rubber interconnections are used and thus the

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†Manfactured by Oliver M. Moe, 1926 Hartford Ave., St. Paul, Minn.

Fig. 2.—*A*, Photograph of the apparatus viewed from the side. The drainage bottle is at the left, air leak indicator in the center, and the Stedman pump on the right. The tube which is attached to the patient's chest catheters is coiled at the bottom of the picture.

B, A photograph of the apparatus from the front. The Stedman pump is seen from the front and the drainage bottle is just behind it. The pressure regulating tube is on the left and the air leak tube on the right. The crossbar at the top is a handle for the apparatus.

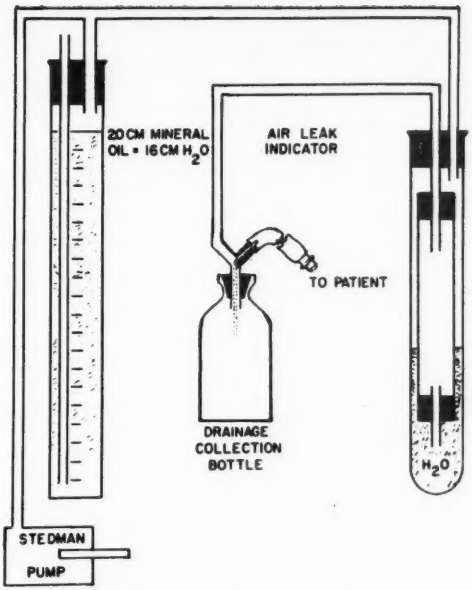
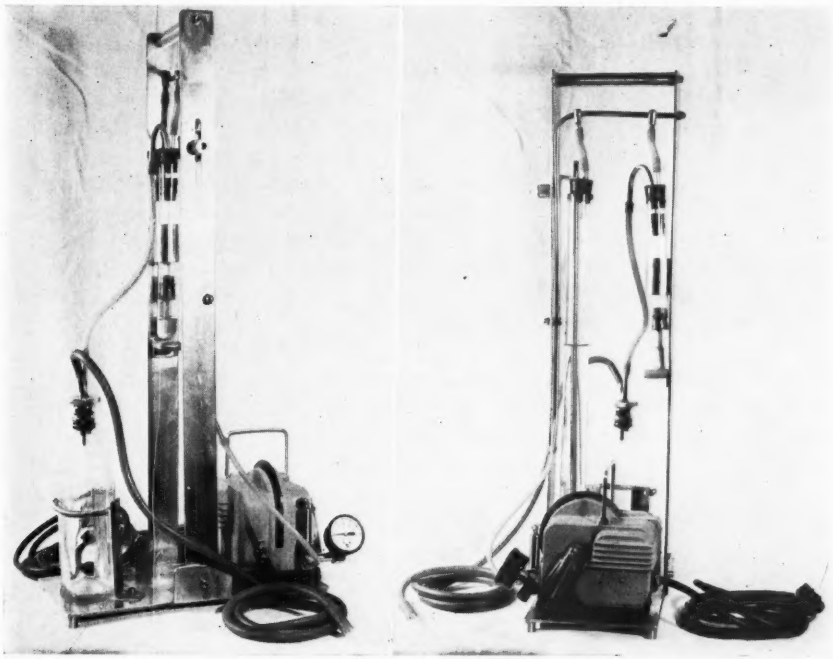


Fig. 1.—A diagram of the suction apparatus. On the left is the Stedman pump and the tube for maintenance of a constant negative pressure. The drainage bottle is in the center and the air leak tube is on the right.



A. *B.*

Fig. 2, *A* and *B.*—*For legend see opposite page.*

danger of kinking is eliminated. The drainage bottle is a standard pharmacist's bottle with graduations on the side which make volume measurements simple and direct. A special stopper makes it possible to empty the bottle quickly and easily. This stopper consists of a rubber cork which is compressed in order to tighten it in the bottle neck by simply turning the knurled top of the stopper.

The apparatus has been used in the postoperative care of pulmonary and cardiac surgery patients and it has proved to be superior to the usual 3-bottle suction apparatus.

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